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Wenche Ann Similä

Health-related quality of life in Young People with Chronic fatigue syndrome/ Myalgic encephalomyelitis

Doctoral thesis

NTNU
Norwegian University of Science and Technology
Thesis for the Degree of
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Department of Clinical and Molecular Medicine

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Helse-relatert livskvalitet blant ungdom med CFS/ME

Kronisk utmattelsessyndrom/myalgisk encefalopati (CFS/ME) er en sykdom som begrenser aktivitetsnivået til de som rammes i betydelig grad. Høyt skolefravær gjør at ungdom med CFS/ME mister viktig lærdom og utvikling sammenlignet med jevnaldrende. Disse ungdommene opplever usikkerhet rundt framtiden og risikerer å bli uføre i ung alder. Målinger av helserelatert livskvalitet har vist at ungdom med CFS/ME scorer lavere enn andre ungdommer. Hovedhensikten med dette prosjektet var å undersøke helserelatert livskvalitet, samt faktorer i skole og hverdagsliv, som kunne være assosiert med helse-relatert livskvalitet blant ungdom med CFS/ME. Først undersøkte vi helserelatert livskvalitet og faktorer i skole og hverdagsliv i en kohort av ungdom med CFS/ME i en tverrsnittstudie. Deretter undersøkte vi erfaringer blant ungdom med CFS/ME i forhold til skole og hverdagsliv i en intervjustudie. Til slutt undersøkte vi lærere, rådgivere og helsesykepleieres erfaringer med å legge til rette skolehverdagen for ungdom med CFS/ME i en intervjustudie.

Først fant vi at ungdom med CFS/ME skåret lavere på livskvalitet enn friske og ungdom med andre kroniske sykdommer, og at kontakt med skole og lærere kunne være assosiert med høyere livskvalitet. Dette kunne handle om at tilrettelegging økte kontakten med skole og lærere og at det igjen førte til høyere livskvalitet. Det kunne også ha sammenheng med at elever som hadde færre helseplager fra CFS/ME klarte å ha mer kontakt med skole og lærere. Videre fant vi at en tilrettelagt plan for undervisning og sosialt liv i skolen kunne øke muligheten for å fortsette skolegangen sammen med jevnaldrende. Mangel på en tilrettelagt plan for undervisning og sosialt liv kunne føre til høyere skolefravær og tap av lærdom, sosial kontakt og utvikling sammen med jevnaldrende. Dette igjen kunne føre til bekymring for framtida og depressive tanker. Til slutt fant vi at lærere, rådgivere og helsesykepleiere hadde erfart at ungdom med CFS/ME mistet tillit til skolen. Det var utfordrende for lærere, rådgivere og helsesykepleiere å forstå elevens behov før diagnose ble satt og før informasjon ble overført fra helsetjenesten. Andre utfordringer var å opprettholde lærer – elev relasjonen og kontinuitet i undervisningen. Som tiltak for å forbedre håndteringen av ungdom med CFS/ME i skolen foreslo lærere, rådgivere og helsesykepleiere følgende; å problematisere skolefravær tidlig; ha et tverrfaglig samarbeid rundt tidlige tiltak; sikre opprettholdelse av lærer – elev relasjonen; og øke kompetansen om CFS/ME i skolen. Dette kunne bidra til å forebygge funksjonstap og skolefravær blant elever med CFS/ME.

Oppsummert så var helserelatert livskvalitet blant ungdom med CFS/ME assosiert med kontakt med lærere og skole. Dette kunne være som følge av tilrettelegging av skolegangen eller grad av helseplager knyttet til CFS/ME. Tverrfaglige strategier for tidlig tilrettelegging av undervisning og sosialt liv i skolen for ungdom med CFS/ME kan bidra til at disse ungdommene deltar i utdanning og sosial utvikling sammen med jevnaldrende. Manglende tilrettelegging av undervisning og sosialt liv i skolen kan føre til tap av utdanning og sosial utvikling sammen med jevnaldrende

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Trondheim 28.04.22,
Wenche Ann Similä

Summary

Background

Chronic fatigue syndrome/Myalgic encephalomyelitis (CFS/ME) is a disease that affects people of all ages. CFS/ME significantly limits the activity level of those affected, including in relation to physical activity, schooling, occupational life and social life. High levels of school absence among young people with CFS/ME result in loss of important learning and social development among peers. As such, there is increasing uncertainty about their future, and personal and socio-economic consequences could put them at risk of becoming disabled at a young age. Measurements of health-related quality of life (HRQoL), including being able to function in school, have shown that young people with CFS/ME score lower than their counterparts without CFS/ME.

Aims

The overall aim of this project was to explore HRQoL among young people with CFS/ME, including the factors associated with HRQoL in relation to school and everyday life. More specifically, the aim was to firstly (Study1) examine HRQoL, including factors that are positively or negatively associated with HRQoL, in a cohort of young people with CFS/ME. Study 1, along with the previous literature, provided the basis for an in-depth study (Study 2) to investigate the positive and negative factors that young people with CFS/ME experience in school and everyday life. Based on the findings from Study 1 and Study 2, a third study (Study 3) was planned to explore teachers', counsellors' and school nurses' experiences with educational and social adaptation at school for young people with CFS/ME.

Method

To explore HRQoL and the factors associated with HRQoL among young people with CFS/ME (Studies 1 & 2), a cross-sectional survey- and interview-based study was conducted. The participants of the cross-sectional study were recruited to participate in the interview study. To explore the experiences of teachers, counsellors and school nurses with education and social adaptions at school for young people with CFS/ME (Study 3), an interview study was conducted with participants recruited among school personnel and school nurses in secondary school (educating students aged 13-16), high school (educating students aged 16-19) and educational psychological services (EPS).

Results

A total of 63 participants were included in the cross-sectional study, 18 of whom participated in the interview study. A total of 12 participants were included in the interview study with the teachers, counsellors and school nurses. In the cross-sectional study (Study 1), young people with CFS/ME scored lower on HRQoL than their counterparts who were healthy or had other chronic diseases. Contact with school and teachers was associated with a higher HRQoL among young people with CFS/ME. This association could be due to that more contact resulted from adaptations of education and social life at school, or that fewer health problems due to CFS/ME had abled the young people to maintain the contact with school and teachers. In Study 2, it was found that an adapted plan for education and social life at school for young people with CFS/ME could increase the possibility of continuing schooling with peers. The lack of an adapted plan for education and social life at school could lead to increased school absence as well as loss of education, social contact and development among peers. Subsequently, this could lead to depressive thoughts and worry about the future. The school personnel and school nurses in Study 3 experienced that young people with CFS/ME lost confidence in school. The challenges experienced by school personnel included (1) understanding students' needs before they received a diagnosis and before school personnel received information from healthcare providers and (2) maintaining the teacher–student relationship and (3) the continuity of teaching. In terms of measures for better management, early problematization of school absence, interdisciplinary collaboration on early measures, ensuring the maintenance of the teacher–student relationship and increasing CFS/ME-related competence in schools were proposed. These measures could contribute to prevent loss of function and school absence among young people with CFS/ME.

Conclusion

HRQoL among young people with CFS/ME was associated with contact with school and teachers, but a causal relationship could not be proven. Interviews with young people with CFS/ME and school personnel suggested that interdisciplinary strategies for early adaptations to education and social life at school for young people with CFS/ME may benefit education and social development among peers for young people with CFS/ME. Lack of educational and social adaptations at school might lead to loss of education, social life and development among peers.

Sammendrag

Bakgrunn

Kronisk utmattelsessyndrom/myalgisk encefalopati (CFS/ME) er en sykdom som begrenser aktivitetsnivået til de som rammes i betydelig grad. Både fysisk aktivitet, skolegang, arbeidsliv og sosialt liv påvirkes. Høyt skolefravær blant ungdom med CFS/ME gjør at de mister viktig lærdom og utvikling sammen med jevnaldrende. Framtiden til disse ungdommene blir usikker, og personlige og samfunnsøkonomiske konsekvenser kan være at de risikerer å bli uføre i ung alder. Målinger av helserelatert livskvalitet og skolefunksjon har vist at ungdom med CFS/ME scorer lavere enn andre ungdommer.

Hensikt

Hovedhensikten med dette prosjektet var å undersøke helserelatert livskvalitet blant ungdom med CFS/ME, og samtidig undersøke faktorer i skole og hverdagsliv som kunne være assosiert med helserelatert livskvalitet. I første studie ble helserelatert livskvalitet og assosierte faktorer undersøkt i en kohort av ungdom med CFS/ME. I Studie 2 ble funn fra Studie 1 og tidligere litteratur brukt som utgangspunkt for tema i en fordypnings studie som undersøkte erfaringer blant ungdom med CFS/ME i forhold til skole og hverdagsliv. På bakgrunn av funn fra de to første studiene ble det planlagt en tredje studie som undersøkte lærere, rådgivere og helsesykepleieres erfaringer med å legge til rette skolehverdagen for ungdom med CFS/ME.

Metode

For å undersøke helserelatert livskvalitet og assosierte faktorer blant ungdom med CFS/ME (Studie 1 & 2), ble det gjennomført en tverrsnittsstudie med spørreskjema, og en intervjustudie. Deltagere fra tverrsnittsstudien ble rekruttert til å delta i intervjustudien. For å undersøke erfaringer med å legge til rette skolehverdagen for ungdom med CFS/ME (Studie 3) ble det gjennomført en intervjustudie med deltagere rekruttert blant personale i ungdomsskole, videregående skole, pedagogisk-psykologisk tjeneste (PPT) og skolehelsetjeneste.

Resultat

Totalt ble 63 deltagere inkludert i tverrsnittsstudien, og 18 av disse deltok i intervjustudien. I intervjustudien med lærere, rådgivere og helsesykepleiere ble 12 deltagere inkludert. I tverrsnittsstudien (Studie 1) skåret ungdom med CFS/ME lavere på livskvalitet enn friske og ungdom med andre kroniske sykdommer. Kontakt med skole og lærere var assosiert med

høyere livskvalitet blant ungdom med CFS/ME. Denne assossiasjonen kunne enten skyldes mer kontakt på grunn av tilrettelegging av utdanning og sosialt liv i skolen, eller at færre helseproblemer fra CFS/ME gjorde det mulig for ungdommene å opprettholde kontakten med skole og lærere. I Studie 2 ble det funnet at en tilrettelagt plan for undervisning og sosialt liv i skolen kunne bidra til å øke muligheten for å fortsette skolegangen sammen med jevnaldrende. Mangel på en tilrettelagt plan for skolegang og sosialt liv, kunne føre til høyere skolefravær og tap av lærdom, sosial kontakt og utvikling sammen med jevnaldrende. Dette igjen kunne føre til bekymring for framtida og depressive tanker. I Studie 3 hadde lærere, rådgivere og helsesykepleiere erfart at ungdom med CFS/ME mistet tillit til skolen. De hadde erfart at det var utfordrende å forstå elevens behov før elevene fikk en diagnose og før lærere, rådgivere og helsesykepleiere fikk informasjon fra helsetjenesten om tilrettelegging av undervisning og sosialt liv i skolen. Andre utfordringer var å opprettholde lærer-elev-relasjonen og kontinuiteten i undervisningen. Som tiltak for å forbedre håndteringen av ungdom med CFS/ME i skolen foreslo lærere, rådgivere og helsesykepleiere følgende; å problematisere skolefravær tidlig, ha et tverrfaglig samarbeid rundt tidlige tiltak, sikre opprettholdelse av lærer-elev relasjonen og øke kompetansen om CFS/ME i skolen. Dette kunne bidra til å forebygge funksjonstap og skolefravær blant elever med CFS/ME.

Konklusjon

Helserelatert livskvalitet blant ungdom med CFS/ME var assosiert med kontakt med skole og lærere, men en årsakssammenheng kunne ikke bevises. Intervju med ungdom med CFS/ME, skolepersonell og helsesykepleiere antyder at tverrfaglige strategier for tidlig tilrettelegging av undervisning og sosialt liv i skolen kan være nyttig for utdanning og sosial utvikling sammen med jevnaldrende for ungdom med CFS/ME. Manglende tilrettelegging av undervisning og sosialt liv i skolen kan føre til tap av utdanning og sosial utvikling sammen med jevnaldrende.

Will the COVID-19 lockdown help us to better understand young people with chronic fatigue syndrome?

We all know the feeling after the COVID-19 lockdown, of not being able to attend school or work, enjoy social life or leisure activities as normal. The familiar everyday life suddenly disappeared, the future became uncertain, and we became concerned for our health.

Imagine being a healthy teenager between the ages of 12-18 with an active everyday life and future dreams, and suddenly experiencing the same losses due to a chronic fatigue syndrome that no one can understand or cure. Your new everyday challenge for months or years is how to cope with the disease. We asked 63 young people with chronic fatigue syndrome about their health-related quality of life measured by physical, social, emotional and school functioning. We also interviewed some of them to explore how they coped with the disease and what they were offered by health care and school facilitation.

Not surprisingly, their function and health-related quality of life were low. Some of them also lost contact with school and friends. Maybe because it was difficult for others to understand what they were going through. Despite this, these young people had a generous attitude and said, “Nor would I have understood it if I had not experienced it”. Maybe the experience with the COVID-19 lockdown can help us understand the loss of everyday life in these young people. In addition, they must cope with activity limitation due to health challenges, which they find frustrating because they wish to come back to normal life as much as we do. «Everything will be okay» after the pandemic, but for these young people the truth is as one of them said “It`s not that I don’t want to, it`s my body telling me I can’t”.

(From my own assignment in communication to the public, May 2020)

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Abbreviations

CFS/ME	Chronic fatigue syndrome/Myalgic Encephalomyelitis
PEM	Postexertional malaise
POTS	Postural orthostatic tachycardia syndrome
HRQoL	Health-Related Quality of Life
ICC	International Consensus Criteria
CCC	Canadian Consensus Criteria
ICD-10	International statistical Classification of Disease and Health Related problems version 10
IOM	Institute of Medicine
WHO	The World Health Organization
NICE	National Institute of Health and Care Excellence
IACFS/ME	International Association for chronic fatigue syndrome/Myalgic encephalomyelitis

List of Papers

Paper I

Wenche Ann Similä, Vidar Halsteinli, Ingrid B. Helland, Christer Suvatne, Hanna Elmi & Torstein Baade Rø. *Health-related quality of life in Norwegian adolescents living with chronic fatigue syndrome.* Health and Quality of Life Outcomes (2020) 18: 170. doi.org/10.1186/s12955-020-01430-z

Paper II

Wenche Ann Similä, Torunn Hatlen Nøst, Ingrid B. Helland & Torstein Baade Rø. *Factors related to educational adaptations and social life at school experienced by young people with CFS/ME: A qualitative study.* BMJ Open (2021) dx.doi.org/10.1136/bmjopen-2021-051094

Paper III

Wenche Ann Similä, Torstein Baade Rø & Torunn Hatlen Nøst. *Experiences among school personnel and school nurses on educational adaptations for students with CFS/ME: A qualitative interview study.* Frontiers in Pediatrics, section Children and Health (2021) doi.org/10.3389/fped.2021.756963

1 Introduction

This project examined health-related quality of life (HRQoL) including the factors associated with it in relation to school and everyday life in young people diagnosed with chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) in Norway. The project was conducted over two time points: 2017/2018 and 2020. This project also examined the experiences of school personnel and school nurses with educational and social adaptations at school for young people with CFS/ME by exploring at single time point: 2020/2021.

The PhD project was motivated by a lack of knowledge about how to improve HRQoL in young people with CFS/ME. The project was prioritized as part of an evaluation project at the Regional Center for Pain and Complex Disorders at St. Olavs hospital, Norway, funded through the government budget of 2017. The project was also funded by the Norwegian University of Science and Technology (NTNU) and St. Olavs hospital. Few studies in Norway have explored the health of young people with CFS/ME, and from clinical practice, we know that there is a lack of knowledge about effective strategies to improve HRQoL in these young people. The overall aim of this project was to explore HRQoL in young people with CFS/ME, including the factors associated with it, in relation to school and everyday life.

The project utilized a combination of quantitative and qualitative methods. Quantitative data were collected cross-sectionally, and qualitative data were collected through one main interview with each participant and additional questions for the young participants with CFS/ME. The quantitative study (Study 1) was designed to examine HRQoL in young people with CFS/ME and factors that are positively or negatively associated with HRQoL. Data were collected through validated questionnaires with the following HRQoL dimensions and symptom measures: (1) physical, psychological, social, psychosocial and school functioning; (2) fatigue and (3) mood and feelings. A disease-related questionnaire that was recently translated into Norwegian added questions about disruption to school activities or performance due to fatigue or cognitive difficulties (4). Dichotomic questions about factors that were received/not received from follow-up in school and healthcare were added to the questionnaire. The first qualitative part of the project (Study 2) was designed as an individual interview study with young people with CFS/ME. Data were collected using a semi-structured interview guide containing questions about the young people's experiences and perspectives regarding the following topics: 1) how they related to their disease; 2) how they experienced others who related to them being diseased with a medical condition and 3) how they

experienced school-related adaptations in relation to their disease. The second qualitative part of the project (Study 3) was designed as an interview study with individual and focus group interviews with teachers, counsellors and school nurses who had been engaged with educational adaptations for young people with CFS/ME. The interview guide was designed to explore the participants' experiences with educational adaptation for young people with CFS/ME and was based on the results from Study 2. Relevant questions included in the guide were "What, in your experience, is challenging for students with CFS/ME concerning the school?" "What experiences do you have from adapting education for these students?" and "How has the COVID-19 pandemic impacted the school day for students with CFS/ME?"

Originally, a third part of the project was designed as a randomized controlled trial with an intervention that included health-promotive dialogues with young people with CFS/ME and measures of HRQoL before, in the middle of and after the intervention. This part of the project was omitted because of inclusion difficulties that were likely due to the COVID-19 pandemic.

The findings presented in this thesis may have useful implications for the management of healthcare, education and social life for young people with CFS/ME. The following paragraphs present definition of CFS/ME, the current understanding of CFS/ME, as well as the history of CFS/ME diagnosis, aetiology and epidemiology. Second is, a presentation on HRQoL definition and how CFS/ME impacts HRQoL in young people. This is followed by an overview of international recommendations for CFS/ME management, including guidelines from the Norwegian National Guide. Finally, findings from previous research on HRQoL in young people with CFS/ME and a discussion on the management of CFS/ME in young people.

1.1 Chronic fatigue syndrome – Definitions and clinical manifestations

The onset of CFS/ME, which is a disabling disease, is either acute or gradual (1, 2). The major symptoms are fatigue (3) accompanied by post-exertional malaise (PEM), which is a worsening of physical and/or cognitive fatigue and other symptoms after normal activity or moderate exertion. Rest or sleep does not relieve the worsening of symptoms, which may persist for hours, days or weeks (3). The presence of PEM is typical and suggested as a core symptom of pediatric CFS/ME (4). General malaise and pain are common (5, 6), and individual variations of additional symptoms include neurological, cardiovascular, gastrointestinal and respiratory manifestations (7, 8). For example, the overlapping syndrome postural orthostatic tachycardia syndrome (POTS) is a symptom present in a CFS/ME subgroup, and the advice is that especially young patients screen for POTS (3, 9). Thus, the identification of heart rate variability has been suggested as important for pediatric CFS/ME evaluation (10). Some of the more unspecific symptoms reported include dizziness, nausea, headache, anorexia and night sweats (3). The total symptoms load impact on health and daily functioning to different degrees in young people with CFS/ME (11, 12). There are no definitive clinical findings or supplementary examinations specific to CFS/ME, therefore the diagnosis is a symptom diagnosis based on a pattern of characteristic symptoms, with separate criteria for pediatric cases (1, 13).

Paediatric cases of CFS/ME are defined by the internationally accepted Jason criteria (14) as:

> Clinically evaluated, unexplained, persistent, or relapsing chronic fatigue over the past 3 months that: a) is not the result of ongoing exertion, b) is not substantially alleviated by rest, c) results in substantial reduction in previous levels of educational, social and personal activities, d) must persist or reoccur for at least three months (14).

The Jason paediatric definition of CFS/ME includes a list of symptoms (presented in Table 1). In the following paragraphs, the history of the diagnosis of CFS/ME is presented along with the development of diagnosis criteria.

Table 1. Symptoms based on the Jason paediatric definition of CFS/ME

	"The concurrent occurrence of the following classic ME/CFS symptoms, which must have persisted or reoccurred during the past three months of illness (symptoms may predate the reported onset of fatigue).
A	Post-exertional malaise and/or post-exertional fatigue. With activity (it need not be strenuous and may include walking up a flight of stairs, using a computer, or reading a book), there must be a loss of physical or mental stamina, rapid/sudden muscle or cognitive fatigability, post-exertional malaise and/or fatigue and a tendency for other associated symptoms within the patient's cluster of symptoms to worsen. The recovery is slow, often taking 24 hours or longer.
B	Unrefreshing sleep or disturbance of sleep quantity or rhythm disturbance. May include prolonged sleep (including frequent naps), disturbed sleep (e.g., inability to fall asleep or early awakening) and/or day/night reversal.
C	Pain (or discomfort) that is often widespread and migratory in nature. At least one symptom from any of the following: Myofascial and/or joint pain (myofascial pain can include deep pain, muscle twitches, or achy and sore muscles. Pain, stiffness or tenderness may occur in any joint but must be present in more than one joint and lacking oedema or other signs of inflammation). Abdominal and/or head pain (may experience eye pain/sensitivity to bright light, stomach pain, nausea, vomiting or chest pain. Headaches often described as localized behind the eyes or in the back of the head. May include headaches localized elsewhere, including migraines).
D	Two or more neurocognitive manifestations: Impaired memory (self-reported or observable disturbance in ability to recall information or events on a short-term basis) Difficulty focusing (disturbed concentration may impair ability to remain on task, screen out extraneous/excessive stimuli in a classroom or focus on reading, computer/work activity or television programs) Difficulty finding the right word Frequently forget what one wanted to say Absent mindedness Slowness of thought Difficulty comprehending information Frequently lose train of thought New trouble with math or other educational subjects
E	At least one symptom from two of the following three categories: 1. Autonomic manifestations: Neurally mediated hypotension, postural orthostatic tachycardia, delayed postural hypotension, palpitations with or without cardiac arrythmias, dizziness, feeling unsteady on feet-disturbed balance, shortness of breath. 2. Neuroendocrine manifestations: recurrent feelings of feverishness and cold extremities, subnormal body temperature and marked diurnal fluctuations, sweating episodes, intolerance of extremes of heat and cold, marked weight change, loss of appetite or abnormal appetite, worsening of symptoms with stress. 3. Immune manifestations: recurrent flu-like symptoms, non-exudative sore or scratchy throat, repeated fevers and sweats, lymph nodes tender to palpitation, -generally minimal swelling noted, new sensitivities to food, odours, or chemicals" (14).
	Exclusionary conditions are also included in the Jason paediatric definition of CFS/ME.

1.2 History of CFS/ME diagnosis

In 1934, myalgic encephalomyelitis (ME) was defined as a disease, but it was firstly named neuromyasthenia (15). While an infectious cause was suspected (16) a psychological explanation was noted (8, 17). In 1955, after an epidemic outbreak of symptoms similar to neuromyasthenia among healthcare providers, the effort to look for cause(s) and treatment increased (18). In 1969, ME was defined by WHO as an organic neurological disorder (19). Nevertheless, in the 1970s it was proposed as a psychosocial phenomenon and renamed "myalgia nervosa" by the European Psychiatry Association. Lacking an organic explanation, this renaming was adopted as a focus, and research into the aetiology remained limited (20).

In 1986, the first diagnostic criteria of ME was presented (1), and shortly after, two cases of the condition ME were related to mononucleosis infection (21). In 1987, a working group in the United States Center for Disease Control (CDC) reached a consensus on "chronic fatigue syndrome" (CFS), and ME/CFS became the international term used for the disease (1, 22). CFS was included as a medically classified disease along with post-viral fatigue syndrome in the International Statistical Classification of Disease and Health Related Problems, version 10 (ICD-10) (19).

Between 1990 and 2003 several criteria for CFS/ME diagnosis were proposed. In 2011-2012, the Fukuda criteria (23) and the Canadian Consensus Criteria (CCC) (24) were suggested as the International Consensus Criteria (ICC) (1, 23, 25, 26). The CCC (24) are more differentiated, and they distinguish patients with greater physical and cognitive functional impairments from those who are depressed (1). In recent years, various recommendations for criteria derived from internationally accepted criteria, such as from the National Institute of Health and Care Excellence Guidelines (NICE) (27) and the Institute of Medicine (IOM) Guidelines (28) – have been used in studies.

The latest criteria suggested in 2015 by the IOM (8), the Systemic Exertion Intolerance Disease (SEID) criteria, are yet to reach a consensus. The Jason criteria used in this study was developed in 2006 and include elements from the internationally accepted Fukuda adult case criteria (23) and follow the recommendations of Reeves et al. (29).

1.3 Aetiology

Before onset, most patients diagnosed with CFS/ME are well functioning and have high levels of HRQoL. Suddenly or gradually, they experience onset of a flu-like illness and no recovery (30-32). There is still uncertainty surrounding what causes CFS/ME, and this is apparent from the large number of causal factors studied (33). The CFS/ME research field has increased steadily over the last 40 years (33), and multiple biological, psychological and/or social factors have been proposed as predisposing, trigger and maintaining for CFS/ME (34).

Contrary to dualistic models in which either biological or psychosocial causes explain an illness (35), the biopsychosocial model (36) has been relevant in several health-care fields, including that of CFS/ME. The biopsychosocial model explains health and illness as resulting from an interaction among biological, psychological and social factors, as illustrated in Figure 1.

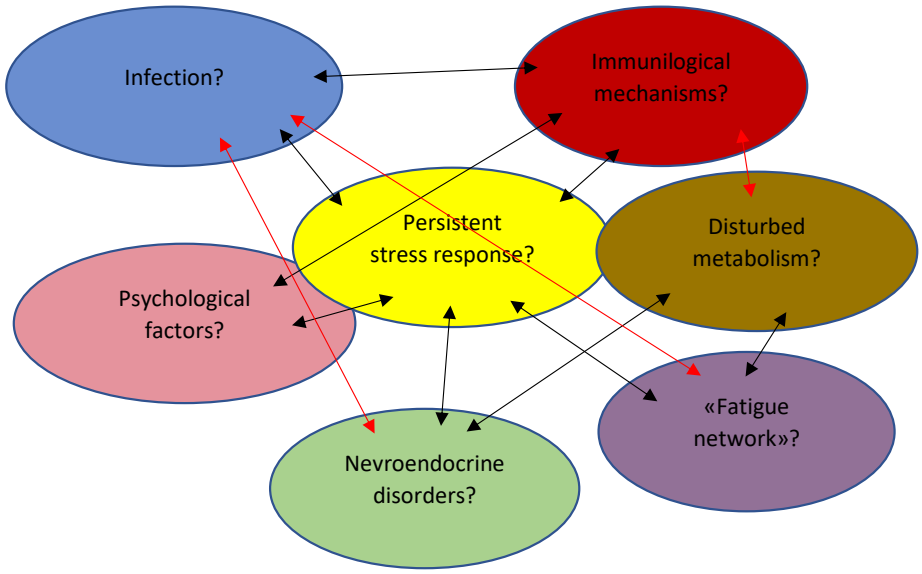


Figure 1. The biopsychosocial perspective on CFS/ME (translated from Norwegian and used with permission from the copyright holders Ingrid B. Helland and Inger Helene Gjone).

However, disagreements about whether CFS/ME is a biological or psychological disease have led to arguments about whether the biopsychosocial model should focus primarily on the psychosocial or the biopsychosocial aspect of the model (35, 37, 38). It was subsequently suggested that CFS/ME should not be defined by physiological, psychological or social

dimensions and that it would be beneficial to CFS/ME patients to take into account the multifactorial and overlapping cause and effect issues (39). Suggestions of combined models for an understanding of CFS/ME were that fatigue was caused by a combination of a comorbid risk, an acute event and patterns of behavioural and biological responses to the event (40); thus, personal responses are what impact the prolonged fatigue (41, 42). Another model included biological changes such as maintaining factors (43).

To improve the understanding of the aetiology CFS/ME, a pathophysiological reference standard based on the connection between the symptomatology and immunological, neuroendocrine and central nervous systems has been suggested (15). Another suggestion is that an integrative profiling of CFS/ME pathogenesis should be developed at the molecular level (44). The most recent studies on causal factors in adults focus on immunology, metabolic dysfunction, psychological reasons and genetics (45-48) Furthermore, in 2021, a map of metabolic phenotypes for adult CFS/ME was presented (46).

It has been pointed out that differences in research designs, number of participants, control groups, diagnostic criteria and methods in general make it difficult to be confident about the studied evidence of CFS/ME (33). The uncertainty surrounding what causes CFS/ME and the lack of consistency about diagnostic criteria have been confusing for medical practitioners (49). It was recently found that between one third and one half of all general practitioners in Great Britain lacked confidence in or did not accept CFS/ME as a diagnosis (50). Therefore, it is important that all research clearly identify diagnostic criteria and treatment before the disease is included in epidemiological and mechanistic studies (51). Thus, differential diagnostic groups can be compared in relation to fatigue level, additional symptoms and treatment (52, 53).

1.4 Epidemiology

Studies on the prevalence of CFS/ME have also been difficult to compare because of differences in the diagnostic criteria of the disease (54, 55). CFS/ME, which is an endemic disorder, is found in all ethnic groups, independent of socioeconomic status, and it is common to find a gender ratio difference of 3:1 or 4:1 between females and males (16, 56, 57). CFS/ME onset is most common in ages 10-19 and 30-39 (58), and the prevalence increases from age 13-18 (59). Nevertheless, children as young as two years of age have also been diagnosed with CFS/ME (60). The prevalence of CFS/ME in young people varies between 0.1- 1.0 %, (55, 61, 62).

The median duration of CFS/ME in adults is approximately seven years (63), and it is more common to experience functional improvements due to the improvement of symptoms than it is with full recovery (64). The duration in young people might be shorter than in adults as it has been found that 75 % of young people regard themselves as having recovered after two to three years (59). In young people, the course of disease is unpredictable, including the possibility of a remission and relapse, and it is common to experience slow improvement over time (3).

To predict disease progression, it has been suggested that factors such as severity at the time of onset and disease management should be examined, since poor management might lead to worse outcomes both in young people (65) and adults (66). Lack of acceptance of CFS/ME has been associated with impaired functioning and increased fatigue in young patients (67), as in adults (68). The disease is not fatal, and cause of death among adult CFS/ME patients is usually linked to another illness, such as cancer, or cardiovascular disease (17, 69), or suicide (70). The following paragraphs presents a definition of HRQoL and an overview of how CFS/ME impacts functioning and HRQoL in young people.

1.5 Health-related quality of life – Definition

The concept of health-related quality of life (HRQoL) implies a broader definition of health, with a focus on physical, psychological and social dimensions of well-being. The concept of HRQoL assesses the impacts of a disease on daily living and functioning (71, 72) and is defined by the World Health Organization (WHO) as "A state of complete physical, mental and social well-being and not merely the absence of disease and infirmity" (73). This means that individual HRQoL might be dependent on life orientation and experienced as high in despite of having a chronic health condition (74). In this, individual, societal and contextual factors are important for the individual experience of HRQoL(75). In young people, especially those with a chronic health condition, the experience of a physical, mental and social well-being might depend on significant adults and the opportunities for facilitations ensuring participation in education and social life among peers (76, 77). The relevance of studying HRQoL in young people with CFS/ME is further addressed in the following paragraphs.

1.6 Impairments in young people with CFS/ME

The impairments experienced by young people with CFS/ME significantly impact their HRQoL (78). It is common for fluctuating fatigue- and individual differences in symptoms

and impairments to impact daily functioning from a mild to very severe degree (3, 11). The division in degrees of CFS/ME was suggested by the ICC (1) and is explained in Table 2:

Physical, cognitive and social functioning are affected:

- In young people, the physical impact causes school absence and, to varying degrees, a lack of participation in sports and leisure activities (79, 80).
- The cognitive impact, concentration and memory impairments functionally disable young people with CFS/ME in school and other activities (81).
- The physical and cognitive impairments cause isolating effects (82) and might increase dependency on parents (78).
- Social implications include reduced functioning leading to loss of, or changes in, relationships with friends and family members, subsequently impacting social development (78, 83).
- The degree of school functioning is individual and depends on, amongst other things, the school's flexibility in educational adaptations (11, 78, 84).
- Depressive thoughts owing to CFS/ME are not unusual among young people, and some develop anxiety (82, 83).

Table 2. Degrees of CFS/ME and associated impairment of function (1).

Degree:	Activity level:	Which means:
Mild	50% reduction in activity	Self-help, can do some housework and manage some school attendance or employment; must regulate leisure and social activities; need to rest some days and at weekends.
Moderate	Mostly housebound	All activity is severely reduced; need to sleep during the day
Severe	Mostly bedbound	Only manage to do activities such as brushing teeth and eating. Many have severe cognitive impairments and are dependent on a wheelchair.
Very severe	Bedbound and dependent on help for physical functions	Need care 24/7; need help with personal care and food; very sensitive to sensory impressions; and some need tube feeding

Depressive symptoms have been found to be related to the severity of the reduction in functioning (85) and a lack of curative treatment of CFS/ME (86, 87). The fear of activities after experiences of PEM negatively impacts school attendance and social adjustment in the life of young people with CFS/ME (88). The development of anxiety has been found to be significantly related to poorer sleep quality in young people with CFS/ME (89), and disrupted sleep could also lead to increased fatigue in CFS/ME patients (90, 91). Figure 2 shows that this can lead to worsening of fatigue.

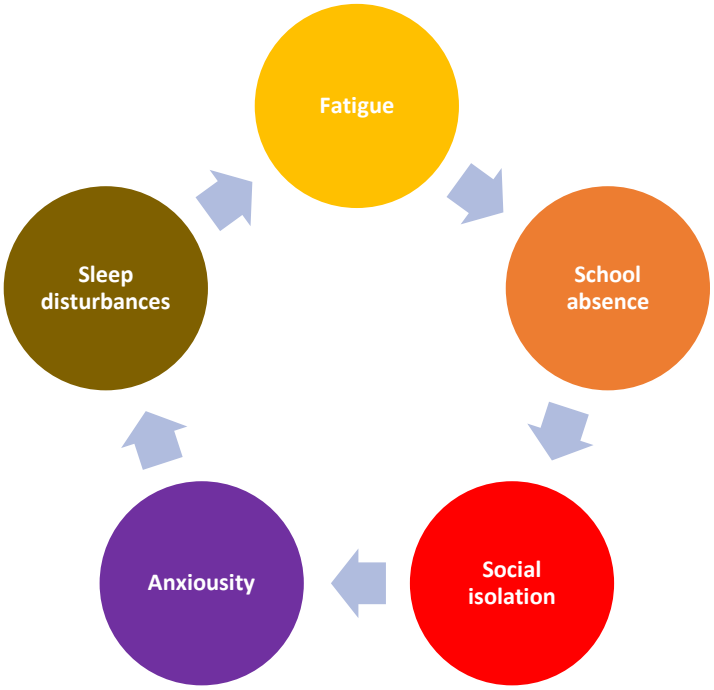


Figure 2. Exemplified path leading to worsening fatigue in young people with CFS/ME.

Since CFS/ME impacts psychological health and could lead to depression or anxiety (92), this might prolong the course of the disease. The impairments young people with CFS/ME experience, occurs in a critical developmental phase of life, the subject of the following paragraphs.

1.6.1 Development in adolescence and young adulthood

According to the WHO, young people cover the age range 10-24 years. Thus, several young people with CFS/ME also encounter the important period of life corresponding to the onset of adolescence, that which is, puberty (93), an important transitional period for physical and

behavioural development (94-96). This development in young people involves significant physical, psychological, cognitive and social changes (97). It is also during this period that young people usually explore a variety of life domains, and the important formation of identity develops through the adoption of values, principles and roles in processes shaped by environmental factors and interaction with peers and significant adults (98-100). Young people with CFS/ME experience reduced functioning and absence from arenas where young people generally develop, potentially causing negative experiences in relation to self-development and the need for strategies that empower the management of self-development (101).

One area where such strategies might be relevant concerns the process of accepting the change in identity in young people with experiences of CFS/ME and, thus, to be able to go on with life (102, 103) which is described regarding adults with CFS/ME (104-106). This might also include body image since this is a domain that impacts identity, especially in adolescents (107, 108), and the risk of obesity that comes with severe CFS/ME might challenge young people's desire to fit in with peers (109).

Another area might be young people's development of independence from parents. Young people usually become better at expressing themselves, which means that advice from adults is not always accepted, rules at home are negotiated, and young people might often think that no one can understand how it is to be them (98, 110). However, young people with CFS/ME have difficulty expressing their disease to others, and they often become more dependent on their parents (78).

Young people do not want to stand out too much from the herd with which they identify themselves (111, 112). How they identify themselves and how they perform compared to their peers impact their dreams and hopes for the future (113). Young people with CFS/ME have the experience of standing still and not being able to participate in life as they watch their peers move on (78).

Young people's ability to think abstractedly and hypothetically about the future is not fully developed until after adolescence; thus, it might be difficult for many young people with CFS/ME to imagine various outcomes of a case or situation (114). This also applies to the ability to think about future responses to different types of behaviour (115, 116). In addition to uncertainties surrounding the course of CFS/ME, this might complicate the ability of young

people with CFS/ME to imagine their future (78), thereby emphasizing the need for young people with CFS/ME to manage their healthcare and school journeys with adequate support.

The following paragraph presents an overview of international recommendations for the management of CFS/ME, including recommendations from the Norwegian National Guide for the Management of CFS/ME.

1.7 Management of CFS/ME

The management of CFS/ME is an individual exercise since the symptoms and severity differ from person to person. Furthermore, as with other chronic diseases, it requires continuing management and periodic re-evaluation (3). Because of the long-term nature of CFS/ME, thorough and repeated assessments of the patient are necessary to decide on course of action (12). It is also important to consider the degree of CFS/ME and the fluctuation of symptoms before a decision on management is made (1).

Following an assessment and diagnosis, specialized healthcare providers furnish CFS/ME-related information to the local healthcare service and schools through interdisciplinary collaboration and individual plans (3). An important focus of healthcare are symptom management and functional improvement through a flexible plan (3). Recommendations for the healthcare management plan for young people with CFS/ME are presented in Figure 3.

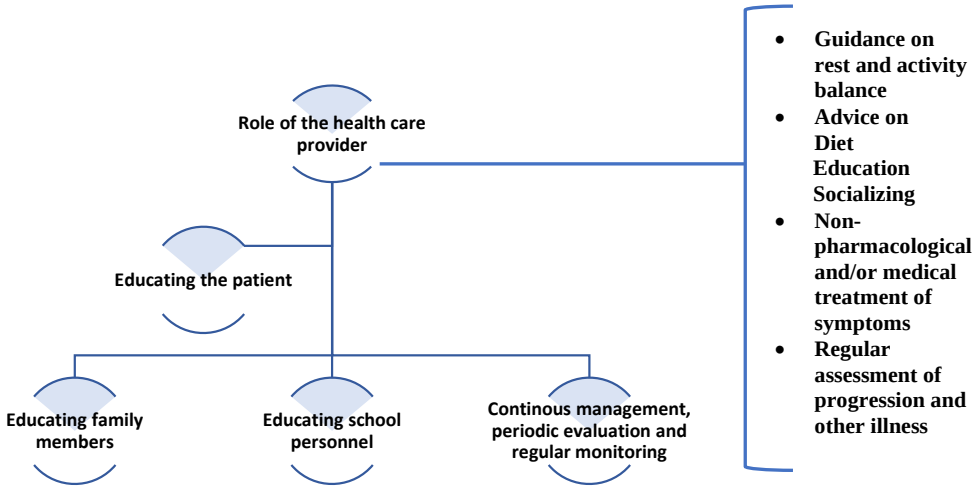


Figure 3. Management of CFS/ME in young people and the role of the healthcare provider (modified from Rowe et al (3)).

Guidelines, especially for Norway, are provided in a national guide from the Norwegian Directorate of Health for assessment, diagnostics, treatment, rehabilitation, nursing and care of patients with CFS/ME and were delivered in 2014/15 (117). The Norwegian national guide for CFS/ME was informed by international research and recommends the internationally accepted Jason paediatric criteria for the assessment and diagnosis of paediatric cases (14). The primary healthcare providers involved in the follow-up of young people with CFS/ME in Norway are usually as follows, bearing in mind that local availability determines who provides healthcare for these young people:

- A general practitioner who is responsible for the follow-up of young people with CFS/ME after a diagnosis from specialist healthcare. The general practitioner's role is to assist with referrals to primary and specialist healthcare services, apply for financial support, take necessary blood samples, check for other illnesses, arrange at-home visits, give advice to the patient and their family and inform them about the individual plan for disease management.
- The occupational therapist maps the daily activities and helps the patient achieve a conscious distribution of energy and balance between activity and rest with a plan of daily activity.
- The physical therapist provides advice about balance between activity and rest and instructions about relaxing exercises, how to improve the quality of rest and sleep and exercise for better balance.
- The nutritionist maps the diet and symptoms to find possible diet changes that can lead to symptom improvement or help with a diet that is better for the patient.
- The social worker provides advice on the legal rights for financial or artificial support as well as help with applications and with mapping the patient's social environment to look for possible resource mobilization initiatives that can assist the family.
- The psychologist offers cognitive therapy to help the patient and their family handle the disease and sends references to rehabilitation services. Controlling for the eventual development of depression or anxiety is also an important part of the role (118).
- Other professions could include a psychiatric nurse who can support with dialogue around psychological reactions to the disease, stress, change of thought and behavioural patterns and consciousness about resources for better mastering. The

nurse is relevant for home care when needed, and environmental workers can render support with social and environmental conditions (117).

Young people with CFS/ME might need guidance to balance activity and rest (3), and it is important to recognize CFS/ME symptoms early in relation to young people's HRQoL (119). To avoid isolation, safeguard development and maintain HRQoL in young people with CFS/ME, special measures for education, social life and leisure activities might be necessary (75). For this, interdisciplinary collaboration on goals and measures and the development of an individual plan will be helpful (120). An interdisciplinary plan can secure a holistic, coordinated and individually adapted healthcare service and strengthen the collaboration around young people with CFS/ME. Interdisciplinary collaboration includes participation from the patient(user), parents, healthcare providers and school personnel (117). Previous research describing HRQoL in young people with CFS/ME has found factors and key themes with the potential to guide collaborative management between healthcare and schools (3), the subject of the following paragraphs.

1.8 Health-related quality of life in young people with CFS/ME

HRQoL has increasingly been considered an important area of research involving young people (121). The advantages of using HRQoL measurements in paediatric healthcare settings are that it facilitates communication between healthcare providers and young people, and HRQoL measurements are helpful in identifying comorbidities and useful for clinical decision-making (122). The following paragraphs present an overview of previous quantitative and qualitative research on HRQoL in young people with CFS/ME.

1.8.1 Quantitative research

Previous quantitative studies on HRQoL in young people with CFS/ME have found significantly lower scores compared to their healthy counterparts and those with other chronic diseases (79, 123-125) (Table 3). HRQoL measures have also shown that school and social functioning are severely impaired (125). HRQoL in young people with CFS/ME also differs from that in adults (5). Factors associated with HRQoL previously found in quantitative research are presented in Table 3.

Table 3. Factors in quantitative research associated with HRQoL in young people with CFS/ME.

ID	Overall factors	HRQoL in young people with CFS/ME is associated with:
YP + P	**School attendance** **Problem solving** **Worry** **Anxiety**	Significantly more illness impairment, especially in school attendance and school issues, than with other chronic diseases. School absence related to reduced physical functioning. Higher levels of worry about illness. Depressive symptoms and being a patient. Anxiety and high levels of worry in relation to illness duration. Low use of problem-solving as a strategy to cope with illness and disability and, rather, the use of emotional regulation to cope with illness, self-criticism or resignation to disability. Criteria: Oxford, NICE, Nor CAPITAL, or not found (123, 126-128)
YP	**Low competency in adolescent domains**	Low competency in areas such as physical functioning, school performance, poorer school attendance, social activities, emotional functioning, romance, participation in recreational activities and general health. High adjustment to adult social standards of behaviour. Normal adjustment to psychosocial self-esteem, social abilities and attentional abilities (85, 125, 129). (No association with duration of illness was found). Criteria: CDC or NICE guidelines
YP	**Social isolation** **Social adjustment** **Depression**	Social isolation and depressive symptoms. Depression and poor social adjustment. Depression related to disability and female sex, higher levels of fatigue, disability, pain and anxiety (130, 131). Criteria: CDC criteria + other patients presenting with fatigue, according to Sharpe et al (132).
YP	**Cardiovascular risk**	Elevated nocturnal heart rate in young people with CFS/ME may pose a cardiovascular risk (133). Criteria: The Royal College of Paediatrics and Child Health

ID = Informant; YP = young person; P = parent

The focus of school experiences among young people with CFS/ME has previously been related mainly to school absence (75). Therefore, the recommendation is to improve assessment methods to understand how CFS/ME impacts school functioning. There is also the criticism that existing patient-reported outcome measures (PROMs) do not capture the outcomes that are important to young people with CFS/ME (134), and thus, there are new measures under development (135). Previous advice also includes that further research should focus on individual differences, a developmental perspective of CFS/ME and interactive

relational factors (136). A list of PROMs used to measure HRQoL and functioning in young people with CFS/ME is presented in Table 4.

Table 4. Patient-reported outcome measurements used to measure HRQoL and functioning in young people with CFS/ME

PROMs used to measure HRQoL in young people with CFS/ME	Generic vs disease specific	**Reported by**	Description
• PedsQL4.0 **(123, 137)**	Generic	YP/P	Overall HRQoL
• CHQ **(125, 138)**	Generic	YP	Overall HRQoL
• SF-36 **(139)**	Generic	YP	Overall HRQoL
• SPPA (not a published manual) (202)	Generic	YP	Psychological adjustment
• YSR **(140)**	Generic	YP	Overall HRQoL
• FDI **(126, 141)**	Generic	YP	Physical functioning
• KidCope **(126, 142)**	Generic	YP	Coping with stressful events
• Things I Worry About Scale **(128, 143)**	Generic	YP	Self-expressed personal and social concerns
• IAS **(126, 144)**	Generic	YP	Fears, attitudes and beliefs
• SCAS **(127)**	Generic	YP	Anxiety
• HADS **(85, 127, 145)**	Generic	YP	Psychological distress
• MFQ **(146)**	Generic	YP	Depressive symptoms
• CDI = Children's Depression Inventory **(130, 131, 147)**	Generic	YP	Depression
• MOHO = The Model of Human Occupation **(148, 149)**		YP	Occupational participation and functioning
• OSA **(125)** YP		YP	
• EQ-5D-5L **(128, 150)**	Generic	YP	Overall HRQoL
• SDQ = Strengths and Difficulties Questionnaire **(128, 151)**.	Generic	YP	Anxiety, depression and worry
• Chronic Fatigue Syndrome Screening Questionnaire (for adults) **(125, 131, 152)**	Disease specific	YP	Adult CFS/ME symptom specific
• DSQ-Ped **(153)**.	Disease specific	YP	CFS/ME symptom specific

PROM= Patient-reported outcome measurement, YP= Young person, P= Parent. PedsQL4.0 = Pediatric Quality of Life Inventory ™ Generic Core scale version 4.0, CHQ = The Child Health Questionnaire, SF-36 = Short Form Healthy Survey, SPPA = Self-Perception Profile for Adolescents, YSR = Youth Self Report, FDI = Functional Disability Inventory, IAS = Illness and Attitudes Scale, SCAS = Spence

Children's Anxiety Scale, HADS = The Hospital Anxiety and Depression Scale, MFQ = The Mood and Feelings Questionnaire, CDI = The Children's depression inventory, MOHO = The Model of Human Occupation, OSA = Occupational Self Assessment, SDQ = Strengths and Difficulties Questionnaire, DSQ-Ped = De Paul Pediatric Health Questionnaire.

1.8.2 Qualitative research

Qualitative studies involving young people with CFS/ME and healthcare providers engaged with CFS/ME, an overview of which is presented in Table 5, has been important in describing complementary information about HRQoL in these young people. Some of these studies have noted that young people with CFS/ME have suffered from several losses related to their impaired functioning (154), with losses and functioning subsequently influenced by management and positive or negative contextual factors (134). By listening to young people's stories about living with CFS/ME, key themes associated with HRQoL have been identified,.

Previous research on HRQoL and associated factors in young people with CFS/ME was found in the following databases: PubMed, Scopus, Cochrane and Google scholar. Keywords in the search for previous studies were chronic fatigue syndrome, CFS, myalgic encephalomyelitis, health, adolescent, adolescence, young adult, young people, school, teacher, student, pupil, university, rehabilitation, management.

Table 5. Identified key themes in qualitative research associated with HRQoL in young people with CFS/ME.

ID	Overall key themes	HRQoL in young people with CFS/ME is associated with:
HP	Symptoms and functioning	Symptoms. Physical function. Participation in school activity and social life. Emotional wellbeing. Help to cope with the condition (75).
YP	Dietary difficulties	Eating difficulties caused by abdominal symptoms. Too fatigued to eat. Changes to taste and smell. Low mood and anxiety that exacerbate eating difficulties. Negative impact on weight (155)
YP	Understanding of CFS/ME	A search for understanding around the illness. Disbelief and distrust from adults. Feelings of being unable to explain the illness. A disease invisible to others. Own and others' understanding of the illness (82, 102, 154).
YP	Competence in adolescent domains	Low perceived competence in specific adolescent domains such as athletic ability, romance and participation in recreational activities. High adjustment to adult social standards of behaviour. Normal adjustment to psychosocial, self-esteem, social and attentional abilities (129).
YP	Individual differences	Varied and flexible illness attributions and coping mechanisms. Individual differences (82, 136).
YP	Loss and adjustments	Experiences of loss and adjustments. Social isolation (82, 102, 154).
YP	Uncertainty and unpredictability	Perceived influence of the illness. Uncertainty. Unpredictability due to reduction in physical and social participation. Symptoms that fluctuate. The body, the illness and the self (82, 102, 134, 154).
YP	Management and contextual factors	Management and positive or negative contextual factors. The relationship between healthcare and school (134).
YP	Peers' relations	Bullying from peers (82)
YP	Acceptance	Disruption from former occupational lives, being on the sidelines of life with peers, the will to take a new body into account and to have hope for a better future (67)
YP	Emotional difficulties	Difficult emotional experiences. The vulnerable self. Feeling locked in and shut out. Impact on emotional well-being. Subsequent depression as a result of CFS/ME (82, 83, 102, 134, 154).
YP	Future prospects	Status post-recovery. Contributions towards recovery. The handling of life and the hope for a better future. The impact on education and career plans from a long-term illness (82, 102, 154, 119)
	Criteria:	NICE, CDC, Fukuda, Nor Capital, Local Clinic assessment criteria or not available.

ID= Informant, HP = Health practitioners, YP = young people

1.9 The management of CFS/ME in young people

Tables 3 and 5 show that since the 1990s, it has been known that HRQoL in young people with CFS/ME is associated with school absence and social isolation and that these young people might develop depression and anxiety as a result. Other factors associated with HRQoL include losses, unpredictability and worry about the future (82, 102, 119, 134, 154). HRQoL and associated factors experienced by young people with CFS/ME are influenced by management and positive or negative contextual factors (134). School attendance, which is the only measure used to assess functioning in young people with CFS/ME, has been criticized, and it has been suggested that assessment of young people with CFS/ME should include a focus on educational challenges in addition to physical, cognitive and emotional challenges (80, 120, 135, 156-158). Furthermore, interdisciplinary collaboration has been pointed out as necessary to provide adequate management for these young people (3, 159).

Managing the education and social life at school of young people with CFS/ME is dependent on various factors (3), especially from the time these young people begin losing function until a diagnosis is set (78). First, it usually takes time before a CFS/ME diagnosis is set (160) and until information from the healthcare services is provided to school personnel. Second, the young people themselves and their parents, teachers, counsellors and school nurses experience uncertainty about the early management of education and social life at school (11, 78, 161, 162). Finally, local and specialist healthcare providers may not have the requisite knowledge to help these young people manage their challenges (3, 50, 163).

In a previous study, the median time for young people with CFS/ME to begin losing function until they got an assessment and diagnosis was found to be 15.5 months (160). Since the onset often follows a period of infection or other bodily stress, it may take time for the young people and their parents to understand that the condition is chronic, as it is for adults with CFS/ME (17, 164). Second, when they seek help from a general practitioner, further assessment may depend on the general practitioner's understanding of CFS/ME (50, 165). Many have felt that they were not believed or understood by their general practitioner, and some have changed general practitioner before they were referred to a specialist (163).

When young people with CFS/ME are referred to a specialist, it usually takes time to get a diagnosis, especially since a CFS/ME diagnosis is set after excluding several other diagnoses (166). This means that young people usually go through a variety of tests before a diagnosis is set. These tests may include physical examinations, laboratory testing, magnetic resonance imaging, psychological and cognitive testing, psychological assessment and school refusal

tests (3). The thorough assessment and diagnostic procedures are exhausting for already fatigued patients, and PEM following assessment is not unusual (3, 166) and negatively impact school attendance in young people with CFS/ME.

From the moment that young people lose function until a specialist assessment, they usually push themselves to go to school, often experiencing worsening conditions (3). How teachers introduce early measures for students with CFS/ME before a diagnosis is set has been poorly explored. Fluctuating school absence due to fatigue and PEM and insecurity among school personnel about the specific needs of students with CFS/ME, might cause delays in introducing adequate adaptations for these young people (167). A recent study found that early measures in school were based on teachers' educational expertise, conversations with students' parents and intuition rather than knowledge about what adaptive measures students with CFS/ME need (161). This might result in a great deal of effort in terms of trying and failing from the period in which students begin to lose function until a diagnosis is set (125). Subsequently, young people with CFS/ME might lack the support they need for academic performance, social contact and development in this period (167), thereby suffering from unnecessary loss of education and social life (102). The subsequent impact on their functioning and HRQoL might lead to a risk of long-term impaired development of cognitive, academic and social skills (168).

Long-term or frequent school absence can also lead to social isolation (167), which can be prolonged into adulthood (169). Thus, school participation might be important for educational, social and emotional development in young people (167). It is in school that young people gain control over their environment and prepare for their independence. If they are unable to attend school, they may feel devalued (170). Furthermore, social and emotional development among young people happens rapidly, and school is the arena for making agreements about leisure time activities with peers (134). Disturbances caused by prolonged or fluctuating school absences make young people with CFS/ME vulnerable and negatively impact their self-esteem (167). Thus, reintegration into school for young people with CFS/ME might be important.

To facilitate reintegration, school personnel might need to be educated about how CFS/ME impact young people's schooling and how individual differences in symptoms and fluctuation impact young people (3, 11). Thus, information from experienced healthcare providers about

the diagnosis, its implications and adaptational needs is important (3). CFS/ME in young people requires a long-term plan for follow-up, and these young people could benefit from facilitations to discuss school-related problems with the healthcare providers who collaborate with their teachers about changes in difficulties, evaluations of adaptational plans and in cases requiring medical attention (3, 167).

Despite the previous knowledge about HRQoL in young people with CFS/ME presented above, little has been explored about the school context for these young people. This gap in the research impacted the choice of the project's study design. Exploring HRQoL among young people with CFS/ME and factors associated with HRQoL in relation to school and everyday life seemed to be a design that could provide insight with the potential to acquire new knowledge to improve strategies for managing CFS/ME and HRQoL in young people.

2 Aims of the Thesis

The overall aim of this project was to explore HRQoL in young people with CFS/ME as well as the factors associated with HRQoL in relation to school and everyday life.

Paper I

The primary aim of this paper was to measure HRQoL in a Norwegian cohort of young people with CFS/ME, while the secondary aim was to identify factors – prior to diagnosis, at the time of diagnosis and after diagnosis – associated with HRQoL.

Paper II

The aim of this paper was to explore factors perceived as positive or negative among young people with CFS/ME in relation to school and everyday life.

Paper III

The aim of this paper was to explore teachers, counsellors and school nurses' experiences regarding educational adaptations for young people with CFS/ME in secondary school (ages 13-16) and high school (ages 16-19).

3 Materials and Methods

3.1 Study design and measures

An explorative design was considered suitable for this project. Exploratory research is used when the problems investigated are not clearly defined (171). As presented in Tables 3 and 5, the problems defining health-related quality of life (HRQoL) in young people with CFS/ME have previously been described in terms of how CFS/ME impacts functioning (3) and the factors associated with functioning and HRQoL. However, factors relating to educational and social adaptations at school have previously not been clearly described, including how they might be positively or negatively associated with HRQoL in young people with CFS/ME.

In unexplored research fields, the observational factor and collecting data directly from those concerned are especially important (171). Therefore, a combined quantitative and qualitative design was relevant for this project (171, 172). It was also crucial to collect data directly from young people with CFS/ME and educational and healthcare providers who have experience with young people with CFS/ME.

Study I was designed cross-sectionally (172) to explore the status of HRQoL in a cohort of young people with CFS/ME and factors with possible positive or negative associations with HRQoL. Previously validated and reliable questionnaires were used for comparability with previous studies of HRQoL in young people. In addition, we used a customized yes/no questionnaire to explore factors associated with HRQoL. Since the participants were fatigued, the home setting was considered most suitable for the self-reporting.

The qualitative part of the study was twofold. Study II was designed as an interview study to explore factors perceived as positive or negative among young people with CFS/ME in relation to school and everyday life. For this purpose, individual in-depth interviews were considered relevant for obtaining detailed information and insight because they do not require many participants, and they can be conducted in natural settings (173, 174). Individual interviews were also relevant in terms of considering the possible worsening of the disease and individual factors such as fatigue and PEM in the participants during the data collection. Thus, the home setting was also considered for the interviews.

Study III was designed as an interview study to explore the experiences of teachers, counsellors and school nurses with educational adaptations for young people with CFS/ME. Focus group interviews were relevant because they leveraged the value of the interaction in

the studied group (175, 176). The inclusion of individual interviews enabled an in-depth study of upcoming themes. Individual interviews were also considered necessary due to the ongoing COVID-19 pandemic.

3.2 Ethical considerations

All studies were conducted in accordance with the Declaration of Helsinki. The studies presented in Papers I and II were approved by the Regional Ethical Committee for Medical and Health Profession Research in South-East Norway (REK 2017/749). The study presented in Paper III was conducted in accordance with and approved by the ethical standards of the Norwegian Centre of Research Data (420197).

The participants were informed about the studies both orally and in writing, and a written consent form was obtained from all of them. Parents signed on behalf of participants below the legal age (16 years). The participants who were also patients were offered supportive healthcare.

Young people suffering from CFS/ME experience fatigue and PEM after normal activity, which is their main symptom (14). This was considered both in the planning of the study design and during the interviews. Ethical consideration regarding interviews with teachers, counsellors and school nurses included awareness about not mentioning students' names or those of participants during the interviews.

3.3 Participants

Papers I and II included participants from two regional hospitals in Norway: Trondheim University Hospital (St. Olavs hospital) and Oslo University Hospital. Eligible participants were recruited from each hospital's patient register of paediatric patients with CFS/ME. This recruitment procedure was approved by the Regional Ethical Committee for Medical and Health Profession Research in South-East Norway (REK 2017/749). The participants received written information by mail and agreed to participate in the quantitative study by returning a signed consent form.

Participants who had replied to the quantitative study and had agreed to be contacted for participation in the interviews were contacted by phone for inclusion. Oral information about the interviews was provided before the participants gave their consent to participate. The phone call also included an agreement about the time of the interview.

Paper III included participants who were employees in secondary schools (educating students aged 13-16) and high schools (educating students aged 16-19) in Mid-Norway. By obtaining consent from municipality directors, eligible participants among teachers, counsellors and school nurses were recruited from three municipalities in Mid-Norway. Written information about the study was sent by e-mail to principals of secondary and high schools and school nurses. Information about the study was also published on Facebook for recruitment purposes. Eligible participants contacted the researcher for registration to participate. All the participants' characteristics are shown in Table 6.

Table 6. Participant characteristics

	Study I	Study II	Study III
N	63	18	12
Gender			
Female	50	13	10
Male	12	5	2
Age	mean (SD)	years	
	18 (2)	13 - 22	
Occupation			
Teacher			6 (1 with leadership)
Counsellor in school			1
Counsellor in EPS			2 (1 previous)
School nurse			4

3.4 Data collection

Quantitative and qualitative data were collected from eighteen of the participants. Other participants only provided quantitative or qualitative data.

Paper I

The data collection for the quantitative study in Paper I took place between August 2017 and January 2018. The participants were asked to partake in a self-reported questionnaire, which was sent to them by mail for completion at home before returning it by mail in pre-addressed and postage-paid envelopes. The questionnaire was extensive and took time to complete. The self-report questionnaire consisted of the following collection of previously validated questionnaires:

Pediatric Quality of Life Inventory ™ Generic Core Scale, version 4.0 (PedsQL4.0)

HRQoL was measured with the Norwegian version of the Pediatric Quality of Life Inventory ™ Generic Core Scales version 4.0 (PedsQL4.0), a 23-item generic questionnaire developed to measure HRQoL in both healthy and acute or chronically ill children and adolescents (177). The PedsQL4.0 scores provide generic sum and subscale scores: physical functioning (8 items) and psychosocial functioning as the total of emotional functioning (5 items), social functioning (5 items) and school functioning (5 items).

The Pediatric Quality of Life Inventory™ Multidimensional Fatigue Scale (PedsQL-MFS)

Fatigue severity was measured with the Pediatric Quality of Life Inventory™ Multidimensional Fatigue Scale (PedsQL-MFS) (178), a generic 18 item scale that provides a total score. The subscale scores measured three domains: general fatigue (6 items), fatigue related to sleep/rest (6 items) and cognitive fatigue (6 items).

The Short Mood and Feelings Questionnaire (SMFQ)

Depressive symptoms were measured with the Short Mood and Feelings Questionnaire (SMFQ), a 13-item self-report-form measure collectively describing depressive symptoms specific to major depression in the DSM-IV (179).

De Paul Pediatric Health Questionnaire (Norwegian version)

The De Paul Pediatric Health Questionnaire (DPHQ-N) for children and adolescents was used to measure 1) demographic data, 2) CFS/ME-related symptoms from the current CFS/ME criteria according to Jason and 3) experiences of disruption in school activities or performance due to fatigue or cognitive difficulties (14, 23, 180).

Additional questions about contact with school personnel and healthcare providers

Information about the participants contact with primary healthcare providers and school personnel was collected in an additional interview with the participants, whereby an interview guide with dichotomic questions was used.

Data from medical records

One paediatrician from each hospital and one psychiatrist from St. Olav's hospital used a semi-structured guide developed by the research group to collect data from the assessment and a diagnostic evaluation of the medical records.

Paper II

For Paper II, data were collected qualitatively between September 2017 and January 2018. A semi-structured interview guide was developed to ensure that the participants talked about their experiences within the same and relevant topics. The interview guide was based on previous knowledge about the perceived challenges facing young people with CFS/ME, especially in relation to education. The first part of the interview guide addressed being fatigued, while the second part addressed healthcare and support from school, family and friends. Some of the central questions included "How has your school curriculum been adjusted and how have the adjustments worked out for you?" and "How do people you relate to perceive your disease?". To avoid the disadvantage of having to travel, which could lead to PEM, the participants were given an opportunity to choose between being interviewed at home or at the hospital and they could bring a parent if they wanted.

In August 2020, an extra phone call was made to the participants to ask two questions about the experiences of school contact and teaching during the COVID-19 pandemic lockdown affecting schools. Notes were taken from the telephone interviews and added to the data from the original interviews. The total impression from the interviews with the participants was that they were all eager to tell their stories, even though the process could make them fatigued and even though there were no advantages for them. Many said that it was important for them to participate because it could give information that could improve the understanding of young people with CFS/ME in the future.

Paper III

The qualitative data collection for Paper III, including the focus group and individual interviews with teachers, counsellors and school nurses, took place between November 2020 and March 2021. A semi-structured interview guide was developed to ensure that the participants talked about topics that were of relevance to the aim of the study. The interview guide included the following questions: "What, in your experience, is challenging for young people with CFS/ME in relation to school?" "What are your experiences with adaptations to the education of these young people?" and "How has the Covid-19 pandemic impacted the school day for young people with CFS/ME?"

The first focus group interview was held face-to-face with the participants, with the researcher and a co-moderator being present. All participants were seated at least one meter apart in line

with the infection protection distance due to the COVID-19 pandemic. The co-moderator took notes during the interview and gave comments about the group interaction right after the interview was completed and the participants had left. The second focus group was held online due to the COVID-19 pandemic. The members of the virtual focus group were seated in the same room at least one meter apart during the interview, while the interviewer met with them virtually.

All the individual interviews in Paper III were performed virtually. The interview guide was edited before the individual interviews based on data from the two focus group interviews, which provided a supplement to the interview guide, with questions on how the school system and other professionals cooperated when planning the education of students with CFS/ME.

The impression of the participants was that they participated because they thought it was important and interesting. One of them also said that he participated because he needed to learn more about young people with CFS/ME, what adaptations they needed at school and what worked.

3.5 Analyses

Paper I

The quantitative analyses were planned in collaboration with the supervisors of this study and were repeatedly discussed during the analysis process. The analysis is presented in Paper I, but some supplementary information about it is presented below.

This was an exploratory study. Therefore, comparisons were made to explore whether various factors relating to the participants contact with the school and healthcare services could be associated with HRQoL. This was performed by exploring many variables from a relatively small group of participants. This revealed some limitations for the analyses, which are further discussed in the methods discussion. During the planning and analysis, there was a realization that many factors with a possible association with HRQoL were not examined in the study, and also that it would be difficult to determine causal relationships, which is discussed in the methods discussion.

Preliminary results from the quantitative study were presented twice in a national forum for researchers connected to all of the Norwegian Regional Centers for Pain, who provided feedback. Preliminary results were also presented at a national conference for CFS/ME research.

All statistical analyses were performed using SPSS, version 23.

Paper II

The qualitative data for Paper II were analysed using a grounded theory approach (181-183). The intention of a grounded theory analysis is to generate or discover a theory regarding a phenomenon related to a particular situation where individuals interact, take action or engage as a response to the phenomenon and the process that follows (182). In this study, the situation was that young people with CFS/ME were unable to participate in school and social life like they used to before they became ill. The analyses explored how the young people with CFS/ME had experienced their interactions with school personnel and healthcare providers as well as actions of these professionals in response to the young people's impaired function from CFS/ME. In analyses using grounded theory as a method, data are continuously analysed before new data are collected in a "zigzag" process, a constant comparative method where the data collection is continuously compared to emerging categories (182). The consciousness regarding the constant comparative process was present during the data collection. Nevertheless, in this study, the constant comparative process was not carried out completely as described in the grounded theory literature (182). Therefore, the analysis was conducted using a grounded theory approach.

Initially, the data were open-coded into seven categories: 1) health, 2) own perception and acceptance of the disease, 3) others' perception and acceptance of the disease, 4) healthcare and support, 5) school adaptations, 6) emotional challenges and 7) loss and sorrow. The open coding showed that there were differences in how the participants had experienced school adaptations. Some had experienced educational adaptations that worked fine, while others had opposite experiences or missed educational adaptations altogether. Educational adaptations were considered the main concern emerging from the data. This was supported by a visual registration of emotionality among the participants.

The second step of the grounded theory approach was to axially code the main concern with the seven categories from the open coding. In axial coding, the young people's own perception and acceptance of the disease, as well as those of the persons with whom they interacted with (category 2 and 3), were considered important in terms of how the need for school adaptations was resolved. This included what actions were taken and by whom. The

main concern as well as categories 2 and 3 were the leading components when the data were described in the third step.

The third step of the grounded theory approach was to selectively code the data into new categories and describe the content in a storyline and a conditional matrix. Following the selective coding, loneliness emerged as the second most important concern among the informants. Contact with school seemed important to resolve the main concerns regarding educational adaptations and loneliness.

Preliminary results were accepted for presentation at the International Association for CFS/ME (IACFS/ME) conference in June 2020, but the conference was cancelled due to the COVID-19 pandemic. The preliminary results were discussed with the supervisors and a research network on patient education and participation at the university. In August 2021, the results were presented on a virtual poster at the IACFS/ME conference.

Paper III

The qualitative data in Paper III were analysed using the principles of Systematic Text Condensation (STC), which is a descriptive thematic cross-case analysis strategy (184). The analyses followed the four steps of STC in an iterative process. All interviews were subsequently transcribed and analysed.

In step 1, all transcripts were read in order to get an overall impression of the data. Analyses of the focus group interviews were initiated before the individual interviews were performed, and it was decided that some questions would be added to the individual interview guide on how the school system and other professionals cooperated when planning the education of students with CFS/ME. The individual interviews were transcribed and analysed in step 1 in a parallel manner to the initiation of STC in step 2.

In step 2, meaningful units from the transcripts were coded and grouped. MindManager 2020, version 20.0.334 and Microsoft ®Excel® for Microsoft 365 MSO (16.0.13127.21656), version 2008, were used as systematization tools during the analyses. Early sorting of the data in MindManager maps was discussed with the supervisors on several occasions. Figure 4 illustrates the early organization of the preliminary categories during this step.

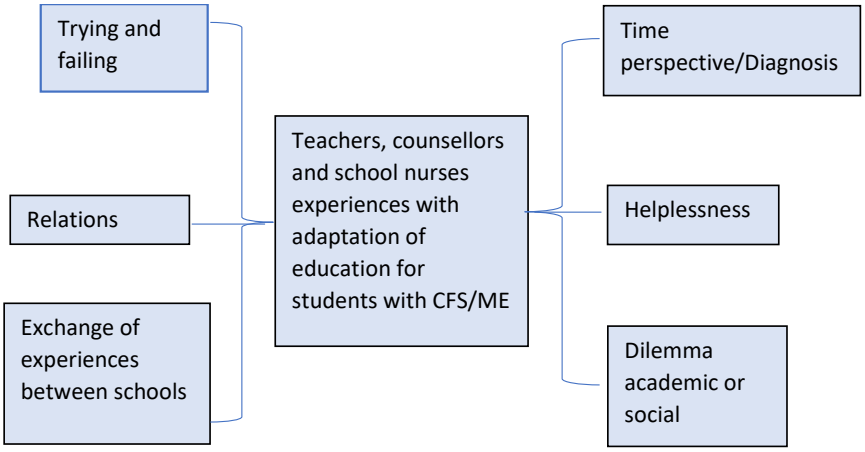


Figure 4. Exemplified MindManager map of preliminary categories in analyses.

Subsequent coding of the data in this step was organized in Microsoft Excel. This enabled a better overview of the categories and an easing of the process for a repetitive check of the results against the original data and transcripts for validation. The preliminary categories were merged into three main categories: *understanding* of the challenges and needs of educational adaptations for young people with CFS/ME; *what happens as the time goes by* with the informants understanding of the student's loss, possibilities to initiate educational adaptations and interdisciplinary collaboration and, finally, the *possibility to prevent losses* and by whom.

The groups of meaningful units were further gathered in five preliminary themes: (1) cognitive and emotional challenges in students with CFS/ME, (2) adaptations of education, (3) social adaptations in school, (4) relations with student and parents and (5) interdisciplinary collaboration.

In step 3, a systematic abstraction of the meaningful units was provided by reducing the content into a condensate that maintained the informants' sayings.

In step 4, the analytical text was written by the first author and discussed repeatedly with the co-authors, and quotes were added. Preliminary results were discussed with a research group on patient education and participation at the university and presented in an interdisciplinary national forum experienced with paediatric CFS/ME patients. The final categories were 1) adaptation of education before the student is diagnosed is challenging, 2) we experience that students lose confidence in school, 3) interdisciplinary collaboration is valuable but sometimes challenging and 4) suggestions on successful adaptations. The results are presented

in Paper III.

4 Results

This chapter presents a summary of the results from the three studies exploring health-related quality of life (HRQoL) and its associated positive or negative factors in young people with CFS/ME in relation to education and social life at school. It first presents a brief summary of each paper, after which detailed results are presented in Papers I-III. Finally, the results are summarized in the last paragraph and Figures 5 and 6.

The overall aim of this project was to explore HRQoL in young people with CFS/ME and the factors associated with HRQoL in relation to school and everyday life at school.

Paper I

The results in Paper I showed that young people with CFS/ME had lower scores on HRQoL than their counterparts who were healthy and had other chronic health conditions. Among several factors explored, contact with teachers and school were positively associated with HRQoL. However, the study could not establish whether contact with school and teachers was the cause of improved HRQoL, if students were more in contact with teachers and school because of fewer health problems and higher HRQoL, or if other factors could explain both increased HRQoL and school attendance.

Paper II

The results in Paper II showed that educational adaptations at school and loneliness were the primary concerns among young people with CFS/ME. For their main concerns to be resolved or taken seriously, they wanted educational and social adaptations that worked and someone to guide them about limitations and possibilities, which, in the participants view, could improve their ability to attend their education. When no one believed them and when they had no guidance on their limitations and experienced improper educational adaptations, they could have a negative course experience, including losses in terms of education and social life. This could lead to depressive thoughts and worry about the future. These findings were new to the field.

Paper III

The results in Paper III showed that it was challenging for teachers, counsellors and school nurses to adapt education for young people with CFS/ME, especially in the period preceding the diagnosis. The challenges were related to school absence, uncertainty about the diagnosis and the students' adaptational needs, and teachers had few meeting points with these students.

This impacted the teacher–student relationship and made it difficult to ensure continuity of education. If they waited for a diagnosis before introducing adaptive measures, the students could lose academic and social development and confidence in school. Suggestions from the teachers, counsellors and school nurses about successful adaptations were as follows: 1) early problematization of school absence and interdisciplinary collaboration with a concrete plan for adaptive measures, 2) focus on teacher–student relationships and 3) increase school competence by exchanging experiences between schools. From the participants perspective, this could contribute to preventing loss and securing educational and social development in students with CFS/ME. Recent experiences with online teaching in the COVID-19 pandemic were potentially useful in developing adaptive measures for young people with CFS/ME. These new findings were provided through an original study design in the study field, namely, to explore how school personnel experienced educational adaptations for young people with CFS/ME. The findings in Studies I-III presented in Papers I-III are illustrated in Figure 5.

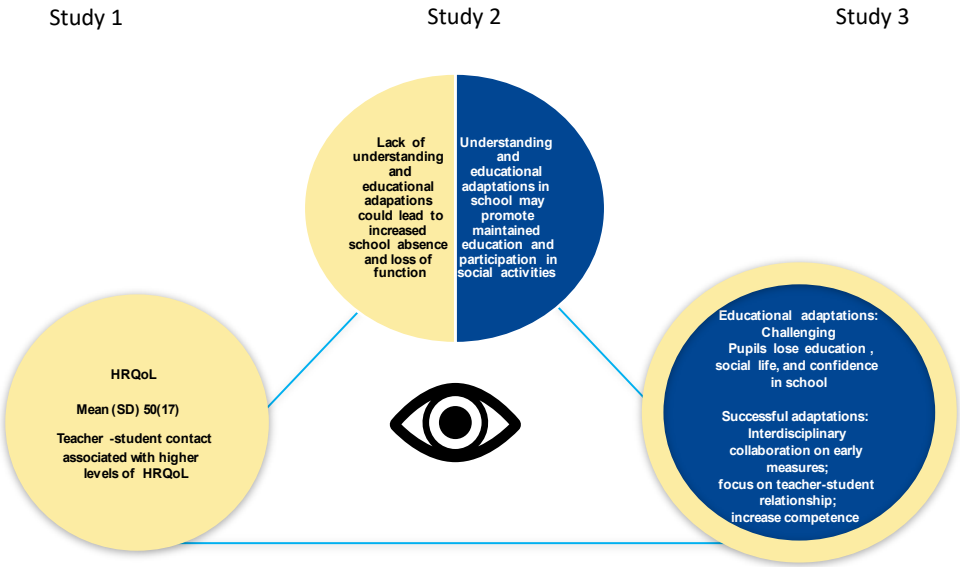


Figure 5. Illustrated summary of the main results in Paper I-III as viewed by the researcher.

4.1 Health-related quality of life and associated factors at school

In sum, Papers I-III found that HRQoL in young people with CFS/ME was severely impaired and that factors relating to education and everyday life at school could have positive or

negative impact on the course of schooling. Positive factors in relation to education and social life at school were suggested as those that potentially could improve the management of education and social life for young people with CFS/ME and, thus, contribute to prevent the loss of academic and social skills.

Some of the positive factors relating to education and social life at school were 1) teacher–student relationship maintenance and 2) early adaptation of education and social life at school, and that this possibly could prevent educational and social losses in students with CFS/ME. To manage this, it was important to 1) problematize school absence early on and not wait for educational adaptations when a diagnosis was set, 2) initiate early interdisciplinary collaboration around adaptive measures and 3) increase competence about CFS/ME in school personnel so that they can become familiar with how to relate to the students and manage adaptive measures and educational continuity. To increase competence, teachers, counsellors and school nurses valued several measures: 1) information from experienced healthcare providers, which they could learn from, 2) exchange of competence between schools and 3) listening to the experiences of young people with CFS/ME who had recovered. These adaptive measures could contribute to prevent educational and social deficits and lost confidence in students with CFS/ME.

Factors that were negatively related to education and social life at school were if whether the students missed the following: 1) having contact with their teacher and the school community, 2) guidance in the balance between activity and rest and 3) educational and social adaptations at school. These factors could lead to the students 1) losing confidence in school and 2) experiencing educational and social development deficits. Possible emotional reactions could include that they 1) became depressed or anxious and 2) worried about the future. However, underlying causes for low HRQoL, such as fluctuating and severe health problems due to CFS/ME or other factors, could also be responsible for the young people being unable to stay in contact with school or teachers despite adaptations of education and social life at school. A summary of factors that were positively or negatively associated with HRQoL is illustrated in Figure 6.

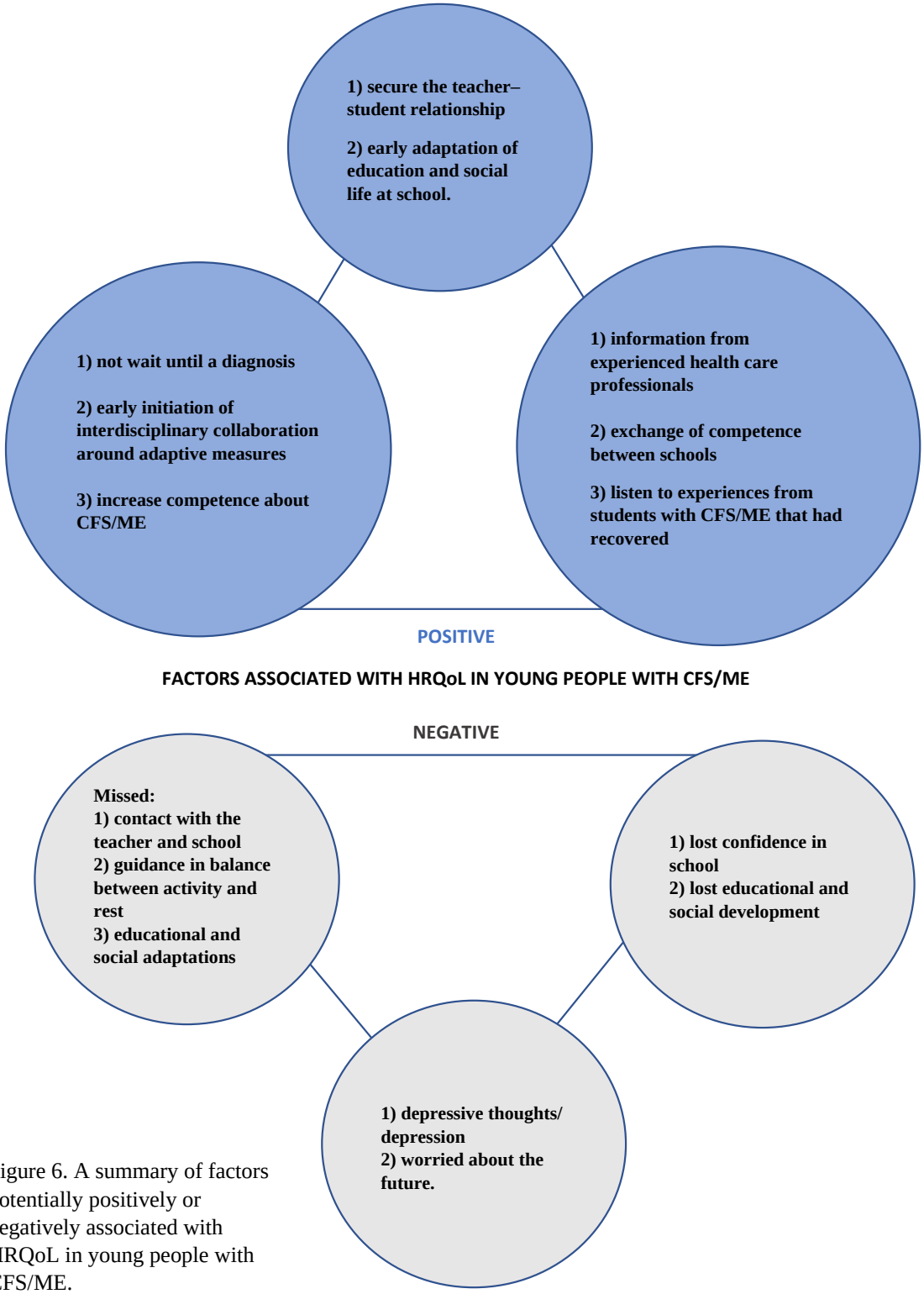


Figure 6. A summary of factors potentially positively or negatively associated with HRQoL in young people with CFS/ME.

5 Methodological Discussion

Consideration of the entire research process, including the initial research question, the choice of design, how the data were collected, how the analyses were performed and what conclusions were drawn is important (173). In quantitative research, such considerations apply to internal and external validity and the reliability of the method and results (185-187). In qualitative research, they apply to relevance, validity, transferability and reflexivity (173, 188).

Decisions made during the research process may affect the results (189-191). Therefore, it is necessary to be aware of factors that may limit the credibility and generalizability of the data. These limiting factors include systematic errors, usually referred to as bias, random errors and generalizability (185, 192). Below, the methodological discussion of this project follows the presentation of the material and methods and starts with reflexivity, followed by a discussion of the design, data collection and analysis.

5.1 Reflexivity

In research, it is important that the researcher exercises objectivity when conducting the research process (193). The researcher's motives, background, perspectives and preliminary hypotheses may impact the research process; therefore, it is important for them to be aware of their own preconceptions (173). Reflexivity may be both challenging and beneficial for the researcher. Challenges may be related to personal matters and the structure of the project, while benefits may include knowledge that secures the ethical treatment of informants and improves the quality of the research generated (194).

My motives for this project were established when I was asked to participate in an exploration of health-related quality of life (HRQoL) in young people with CFS/ME. The Department of Health and Care in Norway had an interest in generating more knowledge about the patient group and had provided funding through cooperation with the Center of Pain and Complex Disorders at St. Olavs hospital. My background with the patient group was approximately one year as a liaison nurse in an interdisciplinary team at St. Olavs hospital that assessed and diagnosed young people with CFS/ME. I had previously worked as a nurse in the adult cardiology field for 17 years. My genuine interest in young people's health led to a change in occupation when I wrote my master's thesis. I chose to immerse myself in the field of health promotion among young people in relation to school health services.

My perspectives as a nurse in this project were holistic. My preconceptions of young people with CFS/ME were that it was important to ascertain their HRQoL and how it could be improved, which meant that we could not just wait to discover the disease aetiology and treatment. In my occupational position, I had experienced that healthcare providers and school personnel wanted to help young people with CFS/ME but that their strategies failed to work. I had experienced that this was challenging for the young people with CFS/ME, their parents, teachers and healthcare providers. With this preconception, I entered the research field with a sense of respect for everything I would encounter.

I had a preliminary hypothesis that health promotion could in some way, help these young people improve their HRQoL. Nevertheless, I did not know the field well and decided to keep an open mind for what I would encounter. Thus, my perspectives were phenomenologically oriented as I aimed to search for the essence of the phenomenon being studied by viewing the objects and events observed from the perspective of how they were experienced by the informants (173). This was a strength in terms of the objectivity of this explorative project. My knowledge from my occupation as a liaison nurse for young people with CFS/ME was a strength in relation to the ethical treatment of the participants. This was because I knew that the participants needed consideration regarding their fatigue and other symptoms during the data collection. My previous experience as a nurse was also a strength in relation to the ethical considerations of the participants' anonymity.

5.2 Study design

The decision regarding the combined quantitative - qualitative design for this project was due to the explorative and observational research question as well as a complex and poorly explored research field (195, 196). A combined quantitative and qualitative approach when studying complex health-related topics is a strength because the complexity of the health-related topic can be explored in detail (197).

Quantitative and qualitative methods can be combined in different ways. Qualitative data can be used to enhance the sensitivity and accuracy of a survey. If quantitative data are collected up front, qualitative studies can be added to explore the meanings and implications of the quantitative findings. A third possibility is to enrich the description of a phenomenon through multiple and diverse observations, which is referred to as triangulation (173, 198). In this project, the quantitative findings were explored up front and further enriched with qualitative data, first, from young people with CFS/ME who also contributed to the quantitative data.

Second, qualitative data from teachers, counsellors and school nurses were added to explore the field from another angle. Thus, the design of this project included triangulation. If triangulation in research increases the understanding of the phenomenon being studied, and various sources agree on the findings, then this strengthens the validity of the study (173). In this project, the qualitative data from the teachers, counsellors and public health nurses increased the understanding of how the main concerns of young people with CFS/ME were experienced and resolved. Studies 2 and 3 found challenges related to resolving the main concern about educational adaptations and social life at school and that how it was resolved could lead to both positive and negative experiences for young people with CFS/ME. Thus, this triangulation strengthened the validity of the study. Nevertheless, one limitation of a combined quantitative and qualitative study may be that if the qualitative data are quantified, this can reduce the transferability of the quantitative data (173). In this study, the qualitative data were not quantified.

5.3 Ethical considerations

Ethical considerations includes the importance of avoiding harm to participants (199). In the quantitative part, Study I, possible harm was decreased by offering the home setting for questionnaires and thus, avoiding the potential worsening of fatigue due to travelling. Nevertheless, there may have been ethical aspect in the home setting that were not observed since the researcher was not physically present. This issue was considered during the planning of the study, but a data protection impact assessment did not find this to be of considerable harm to the participants. Harm was also avoided by choosing a cross-sectional study design and explore relevant factors retrospectively and thus, not deliberately expose the participants to an agent (198).

Ethical challenges in qualitative data collection include maintaining dignity, privacy and confidentiality and avoiding harm (200). Special attention was given to the young people with CFS/ME who participated in the interviews in Study 2. First, they were offered interviews at home to avoid the potential worsening due to travelling. Second, during the interviews, they were told that if they wanted a break, they could stop the interview. This was intended to decrease the possible experience of fatigue during and after the interviews. One participant said that she started feeling fatigued towards the end of the interview but wanted to complete it. Some participants said that they knew that they would be fatigued; however, because they thought it was important to participate, they chose to endure PEM. During one interview, I

experienced that I became emotional myself and expressed this to the informant, who responded by saying that it was okay and that it proved that she was being taken seriously. Ethical considerations during the focus groups in Study 3, were aimed at ensuring anonymity, confidentiality and avoidance of risk of harm (201). In Study 3, the participants were informed before the interview that they could not mention the names of students or focus group participants during the interview. Anonymization in the interview transcript was also maintained. All three studies were performed according to the Declaration of Helsinki.

5.4 Participants

The participants for Studies 1 and 2 were young people with CFS/ME from different parts of Norway. Those for Study 3 were teachers, counsellors and school nurses in secondary and high school with experience of young people with CFS/ME.

To increase reliability, it was important that the selection of participants met sample size criteria (174) and that the participants came from different school communities. A valid sample for a cross-sectional study can be made by inviting participants from a relevant health register. One limitation is that this is not necessarily an accurate reflection of the general population. If some individuals in the study have favourable outcomes compared to those who avoid participation, the results will not reflect the general population and will bias the study (202). This selection bias is referred to as the Neuman bias (203).

The young people with CFS/ME in Studies 1 and 2 were invited based on relevant health registers in two different hospitals, thereby more strongly reflecting the general population. One limitation was that informants with the most severe degree of CFS/ME most likely missed out on participating in the study. Some participated in the quantitative data collection but not the interviews. In the in-depth interviews with the young people with CFS/ME, it was possible to include participants until saturation was achieved, which was a strength of the data collection. For the focus group and individual interviews with the teachers, counsellors and school nurses in Study 3, the inclusion of the participants was difficult, possibly due to the COVID-19 pandemic. The aim was to include 16-20 participants, but 12 were included. Nevertheless, the two focus groups consisted of four participants each, which was the lowest number required for a focus group and, therefore, was valid (175). Four additional individual interviews made it possible to conduct a deeper exploration of the topics appearing in the focus group interviews, thereby strengthening the data.

One limitation regarding the small number of participants in the focus groups may be that the study was unable to detect relevant aspects. The participants in Study 3 were engaged in the phenomenon under study, and this strengthened the generation of data. However, this might not be the same for all teachers, counsellors and school nurses.

In qualitative studies, collecting data until saturation is achieved increases the informational power of the data (204). If a study aims to be broad, the number of participants must be higher (204). In an explorative research design, the aim is to present selected patterns of the phenomenon, and therefore, it is not necessary to cover the whole range of the field (204). In Studies 2 and 3, the invited participants covered the range of the research questions since they had experience with CFS/ME and had knowledge about how CFS/ME impacted the functioning of young people.

5.5 Data collection

The data in this project were collected cross-sectionally (172) and with interviews (181). The main aim of the cross-sectional study was to measure HRQoL in a cohort of young people with CFS/ME, and in addition explore factors associated with HRQoL. A cross-sectional study design is well suited to find factors associated with HRQoL. Cross-sectional studies can be used to infer causation by exploring whether the informants have been exposed to a relevant agent and whether they have an outcome of interest (198). One strength of such studies is the benefit of ethical principles, since the participants are not deliberately exposed to the agent (198). In this study, the outcome, HRQoL and its associated factors were explored in relation to the participants' previous experiences. Therefore, there was no deliberate exposure to an agent.

In a cross-sectional study, which is observational, it can be challenging to infer a causal relationship between the variables (205, 206). In this study, to infer causal relationship between HRQoL and other factors, previous knowledge about factors of importance to HRQoL in young people with CFS/ME could have been considered more thoroughly before a decision was made about relevant factors. Thus, the variables included might have been different. A conclusion, that increased contact with school and teachers led to higher HRQoL could be tempting to make on the basis of the positive association between these factors. However, one has to consider the possibility of a reverse causation between HRQoL and the factors experienced by the participants from their past (205, 206). Maybe a higher HRQoL could have caused that contact with school and teachers was maintained.

Additional considerations of confounders when considering causal relationships is also important in a cross-sectional study (172, 205). In Study 1, confounders could for example have been that communication with the teacher could depend on chemistry or understanding between the teacher and the student or parents, and thus, this could have led to less or more contact. Another confounder could for example have been that grandparents were available to drive the student to school, and that this led to more contact with school and teachers. A third confounder in Study 1 could be that participation in leisure activities were dependent on availability and not directly on health problems.

To remove the possibility of a reverse causation and possible confounders in a cross-sectional study, it is necessary to investigate possible causal relationships in separate analysis beforehand (205, 207, 208). Also, a narrower decision about time limitations for the course of disease before inclusion of participants could have been beneficial to reduce the possibility of exposure to confounders because causalities in a cross-sectional study may be unclear if the exposure has developed over time (202). Such beforehand analyses and inclusion criteria were not provided for Study 1. Thus, the association between HRQoL and the factors could have some limitations to the study regarding conclusions on whether factors had a causal significance to HRQoL. Therefore, the probability of associations between HRQoL and factors were more plausible (213).

To explore causal relationships, the findings in this cross-sectional observational study was instead further explored with in-depth interviews with young people with CFS/ME and with school personnel and school nurses experienced with CFS/ME in young people. It was a strength in terms of validity that data were collected from teachers, counsellors and school nurses, in addition to that from young people with CFS/ME, from different school communities. This somewhat increased the validity of the representativeness of the data for the group of participants who attended school.

5.5.1 Quantitative data collection

The strengths of a quantitative survey, as in Study 1, are the time efficiency, cost-effectiveness, systematic data collection, objectivity and that a large sample size is possible (209). The reliability of a quantitative survey is dependent on the structure of the survey and the quality of the replies (187). The reliability of a questionnaire refers to the consistency of

the measurement, that the items measure one construct and that repeated measures are stable (187).

The questionnaires used as tools for the data collection are important for the validity and reliability of the data collected (177). The face validity of a questionnaire refers to the extent to which it is subjectively viewed to measure what it is supposed to. The internal validity refers to whether the tool objectively measures what it is designed for. External validity refers to the generalizability of the collected data to other populations, settings and times (185). One limitations of a quantitative survey is a low response rate. Factors that may bias the data collection are recall bias and missing data (210, 211). A quantitative survey may also be rigid since it does not involve environmental, emotional, or behavioural factors regarding the respondents (212).

In Study 1, the validated questionnaires substantiated the validity and reliability of the findings (137, 213, 214). The PedsQL 4.0 has previously been used to measure HRQoL in both healthy and chronically ill young people, including young people with CFS/ME (123, 215, 216). Thus, one strength of the study is that it was comparable to previous research. The questionnaires sought data from the near past, which reduced the risk of recall bias (198). Nevertheless, some of the additional questions about the adaptations experienced by the informants and who supported them from healthcare and school were related to the total course of the disease, which caused a risk of recall bias.

By deciding that the home setting for the completion of the questionnaire was best for the participants, there was a risk that they would be assisted with completing the survey by family members. This was also expected since the participants were fatigued young people, most of whom lived with their parents and siblings. A strength related to this was that the questionnaires were completed and returned. A limitation may be that the parents or siblings could have influenced some of the answers. The response rate from the invited participants was 37,5 %, with a limitation being that the total sample size was relatively small.

5.5.2 Qualitative data collection

In qualitative data collection, systematic and reflective processes are important to provide relevance and validity, including transferability and reflexivity as overall standards (173). The relevance of a qualitative study means that participants and observations are relevant to the research question. Reliability in qualitative data collection relies on the relevance of tools and

the selected participants (217). Validity refers to principles such as triangulation and clear details regarding the methods of data collection. Triangulation includes informants who cover various aspects of the phenomenon, thereby increasing the validity of the data collection (198). An advantage of a qualitative data collection is that the sample size can be relatively small (209). A disadvantage is that qualitative data may be more expensive to collect than quantitative data because it is time consuming (209). Bias in qualitative data collection may be related to the participants' recall and the researcher's ability to focus on relevant questions to answer the research question (217).

In Studies 2 and 3, it was relevant to develop interview guides with questions aimed at answering the research question for each study. The development of the interview guide in Study 2 was guided by previous literature and clinical experiences with young people with CFS/ME, which increased the validity of the study (173). The interview guide for Study 3 was based on literature (78), the experiences of young people with CFS/ME (preliminary findings from Paper II) and input from teachers with experience with young people with CFS/ME.

Since an explorative design was chosen for this study, open-ended questions in semi-structured interview guides were relevant. Some strengths of an in-depth interview guide are the possibility of regulation in order to get more detailed information, the requirements of few participants, and it may occur in formal settings (174). A limitation may be that it is viewed as incapable of being generalized (218). Another strength of interview studies is the possibility to observe the informants during the data collection. This is a strength in qualitative data collection as participants' expressions during the interview can complement the impression of the topics being discussed (217). For example, several of the participants in Study 2 became emotional when they talked about educational adaptations and loss of education. Another strength is that, during the interviews, it was possible to explore more deeply new and relevant topics that emerged during the data collection. Since factors from the participants' daily life during the course of the disease were explored, there was a chance of recall bias among the participants (210). The participants in Study 2 also had challenges with memory and concentration due to CFS/ME. This may have limited the reliability of the data.

Especially for the focus group interviews in Study 3, the costs were lower than for the individual interviews due to several participants in one interview, which was a strength. Another strength was that by interviewing different profession's, the "elephant" could be

viewed from different angles (173). It was also a strength that the group dynamic may have elicited more data from the participants (175, 176). A limitation may be that the group dynamic limited the data collection because some of the teachers may have been influenced by one leader who participated in the focus group. Other limitations could have been difficulties to moderate the group to focus on the relevant topics, although this was not a problem for Study 3.

5.6 Analyses

5.6.1 Quantitative analyses

All the participants answered the HRQoL measure, the PedsQL 4.0. The questionnaires were checked for missing data, and all replies were included in the HRQoL analyses. In cases of missing data, imputation of the mean of the completed items was considered the most unbiased and precise method recommended for PedsQL 4.0 (137, 219). Eight of the participants did not answer the school functioning part and explained that they did not go to school. Therefore, imputation was considered but deemed irrelevant. According to the scaling and scoring guide for the PedsQL, it could be computed if at least 50 % of the items were completed. All eight participants had completed more than 50% of the scale.

The factors explored in association to HRQoL were incomplete in some cases. This was due to lost-to-follow-up bias in the additional telephone interview, recall bias or that some of the factors collected from medical records were unavailable. One example was that data on school attendance before diagnosis lacked in some medical records. Imputation was not performed to unbiase these variables. Thus, to avoid bias, participants with incomplete data were excluded from the related bivariate analyses.

A calculation of the sample size in a quantitative study is necessary to predict the strength of the calculations in the analyses (220). In Study 1, a sample size calculation was performed for the quantitative part of the study. The calculation aimed for a sample size of 75 participants to be able to compare the tertial with the highest scores on HRQoL against the tertial with the lowest scores to explore eventual correlations with associated factors. The final sample consisted of 63 participants who completed the HRQoL survey, and 48 participants who reported all factors explored. This was a small sample size, which implied that the results from the analyses were interpreted conservatively. The analyses of the large number of factors from a small number of participants were performed with two-sided independent samples t-

tests to find variables of significance to HRQoL (137, 200). We chose not to make correct for multiple testing of factors as the study was exploratory. Significant associations were further explored in a linear regression analysis (137).

One of the study limitations was the possibility of missing significant associations between HRQoL and the various factors before, at the time of or after the diagnosis due to a small sample size. A multiple regression analysis adjusted for confounders in the model. With a larger sample size, we could also have adjusted for potential confounding from other variables (221). Since bivariate analyses were not corrected for multiple testing, thus, the results should be cautiously interpreted. HRQoL is complex and dependent on several environmental, social and psychosocial factors (222). An analysis plan made before inclusion and an improved design of the study could have enhanced the ability to establish causal factors explaining HRQoL. Instead, we added qualitative analyses to explore the topic in a more in-depth manner.

5.6.2 Qualitative analyses

The validity of qualitative analysis has to do with respondent validation, clear detailing of methods of analysis, attention to negative cases, reflexivity, triangulation and fair dealing (173). The sample size in qualitative interview studies with open-ended questions needs to be as large as the saturation requires (174). In Study 2, the sample size was considered large enough when saturation was achieved (174). This was one of the strengths of the study. For the focus groups, the sample size achieved was the minimum number of participants required (174, 220). This may be a limitation of the results; however, one strength of the analysis was that the focus group interviews were complemented by individual interviews.

In qualitative analysis, the researcher is the tool, and data are analysed through the lenses of the researcher (173, 217). It is important to conduct verifications with other researchers to provide guidance at various stages of the analysis process, including the data interpretation and reporting (173). This triangulation was provided for Studies 2 and 3.

The analysis of the qualitative data starts with the transcripts. A thorough analysis was performed by reading the transcripts to get an overview of the data collected, organizing it, breaking it into meaningful units, synthesizing it, searching for patterns to discover what is important and deciding what to tell in the results (184). Reflection on proximity and distance to the data is also important during the analysis (223, 224). The results must be written in the

sense of content (173, 184). A strength in qualitative analyses is the in-depth information that is available on the topic being explored (173). One of the strengths in the analysis of data from focus group interviews is the possibility of exploring several voices and different angles relevant to the phenomenon (173). One limitation of qualitative analyses may be the challenges regarding the generalizability of the results and the researcher's lenses while analysing the data (173). The validity of qualitative analyses relates to the strengths of new knowledge from an unexplored research field (209).

The individuals in Study 2 talked about similar experiences, despite belonging to distant school communities. This gave validity to the analyses and the transferability to populations other than the participants themselves. The validity of the focus group analyses was related to the previously unexplored field in schools where different professions cooperated around young people with CFS/ME. The data provided insights into the system around the young people with CFS/ME, both in schools and in the interdisciplinary cooperation with healthcare providers. It was possible to identify challenges and what caused them. It was also possible to gain insight into possible available resources and those that were lacking. An analysis of the data from the interviews was informed by a grounded theory approach (181, 182) and STC (184).

5.6.3 Grounded theory approach

Through open, axial and selective coding, concepts were examined across their properties and dimensions. The concepts were integrated into a core category that developed during the analyses. Grounded theory originates from sociology and symbolic interactionism, which posits that meaning is negotiated and understood through interactions with others in social processes (225, 226). To answer the research question of Study 2, it was necessary to analyse the core category - educational adaptations at school in relation to the participants' experiences with interaction at school. To develop an explanatory theory of the phenomenon, it was important to consider the participants' experience with social process at school since the phenomenon usually took place in the school environment (227). The social interactions that took place at school were deemed important in relation to positive and negative factors in educational and social adaptation at school for young people with CFS/ME.

5.6.4 Systematic text condensation (184)

The four-step analysis of STC provides a structure suitable for presenting experiences as expressed by the participants. The method requires that analyses are provided with proximity

to the original data (184). The objectivity of the researcher is important (173). STC was considered a suitable method for the unexplored field of Study 3. The variety of professionals interviewed provided multiple viewpoints in the analysis and a triangulation that strengthened the validity of the findings. Another strength was that the analyses were verified by other researchers, both with and without experiences from the field. A possible bias could have involved whether the registered participants were only those with specific experiences.

5.7 Transferability

Transferability is possible with an adequate and sufficiently varied sample, as well as with considerations about whom and what the findings concern (173). Since the participants in Studies 1 and 2 attended different schools, this could improve the validity and transferability of the data, as common experiences would increase the reliability of the findings. A limitation was that the participants may have been influenced by environmental factors, for example knowledge acquired about the topic discussed may have influenced their answers. The participants in Study 3 came from six different schools and shared common experiences and focused on young people with CFS/ME and the challenges they experience (Study 2). This increased the transferability of the findings to other schools.

5.8 Limitations due to funding

Finally, the funding of the study allocated through the government budget was set before the project was planned, and it was limited in amount and time. This funding made it possible to plan the first part of the project, include participants for Studies 1 and 2, collect data, begin the analyses from the quantitative data and write a report with preliminary findings. Supplementary funding, provided by NTNU, was necessary to complete the analyses and produce articles. The funding from NTNU was allocated as a PhD grant and, therefore, provided an opportunity to plan and conduct Study 3. The available funding did impact research performance, size and efficiency (228). If this study had unlimited resources, a better choice of study could have been a prospective randomized study exploring educational adaptations and social life at school among young people with CFS/ME and how interdisciplinary collaboration around these young people was provided (229). This could have enhanced the internal and external validity, transferability and reliability of the study.

6 Discussion of the Main Findings

The data used in this project were collected from young people with CFS/ME as well as teachers, counsellors and school nurses who had previous and/or current experiences of educational adaptation for young people with CFS/ME. The findings of each study are discussed specifically in Papers I-III. In the following paragraphs, the combined findings are discussed with respect to the overall aim of the project, namely, to explore health-related quality of life (HRQoL) among young people with CFS/ME, including the factors associated with HRQoL in relation to school and everyday life. The findings are discussed in the context of their implications and whether they provide new insight (173).

The results presented in Papers I-III confirmed several previous findings: that HRQoL in young people with CFS/ME is severely impaired (79, 124, 125); that young people with CFS/ME are marred by losses in education and social life, which is one potential reason for their severely impaired HRQoL (82, 154), and that school personnel find it challenging to adapt educational practice before they receive information from healthcare providers. Thus, early measures for young people with CFS/ME might not necessarily be based on sound scientific and practical advice (3, 161). The following paragraphs present a discussion on the most important findings of this study. The first, is that young people with CFS/ME experienced contextual differences in resolving their challenges pertaining to education and social life at school and that this could lead to a positive or negative schooling experience. The second, is that the deficits in education and social life at school faced by young people with CFS/ME were associated with their HRQoL. Finally, teachers, counsellors and school nurses suggested measures aimed at preventing some of the losses faced by young people with CFS/ME.

6.1 Contextual factors influencing positive/negative courses

Frequent contact with teachers and school was found to be positively associated with HRQoL in young people with CFS/ME (Paper I). However, a causal relationship between frequent contact with teachers and school and HRQoL was not established. HRQoL could either cause or be the result of contact with teachers and school, and fewer health problems could impact both HRQoL and school contact. Thus, this along with degree of CFS/ME should be explored in future studies. From in-depth interviews, it was found that when teacher contact and adequate educational and social adaptations at school were secured, young people with CFS/ME could have a positive educational experience (Paper II). In line with previous

findings, this could indicate that adequate educational adaptations might enable young people with CFS/ME to keep up with peers (3). Thus, this add to the findings from Paper I and suggest that adaptations at school can increase HRQoL in young people with CFS/ME. Conversely, if these measures were lacking, young people with CFS/ME could have a negative experience in terms of worsening of symptoms, increased school absence and worry about the future (Paper II). This corroborated previous findings that young people with CFS/ME might struggle with adaptations and, thus, experience uncertainty and unpredictability in education and social life (102, 154). A new finding was that teachers, counsellors and school nurses confirmed that young people with CFS/ME could suffer from deficits in educational and social life at school due to disease-related challenges and that, without adequate adaptations, they could lose confidence in school (Paper III). This points to the need to improve the management of education and social life at school for young people with CFS/ME.

Young people with CFS/ME experienced that people's perception and acceptance of their disease were important for resolving educational and social challenges (Paper II). This is in line with previous literature reports, that the management of education for young people with chronic health conditions depends on attitudes in significant adults, that is, school personnel and healthcare providers (167). A positive factor for young people with CFS/ME was being taken seriously by their teachers about their challenges (Paper II). Conversely, experiences of disbelief and being overlooked by their teachers, especially early in the course of the disease, could have negative impacts on education (Paper II). This is in line with the previous finding, that young people with CFS/ME might find it difficult to explain their disease to others and, thus, may not receive the educational and social adaptations they need (82, 102, 154).

The teachers, however, described that it was difficult to understand the students' needs before being informed by healthcare providers, and thus, it was challenging to adapt education for young people with CFS/ME at the beginning (Paper III). This is in line with previous findings regarding challenges teachers face when they adapt education for these young people (161). This indicates that there is a need to increase knowledge about CFS/ME among school personnel.

Young people with CFS/ME wanted to attend school, but they were not always able to (Paper II). Fluctuating and long-lasting school absences impaired the teacher–student relationship and made it difficult for teachers to provide continuity of education for young people with

CFS/ME (Paper III). In line with previous findings, this confirmed that fluctuating school attendance over months or years among young people with CFS/ME require special attention towards educational and social adaptations (3, 230). However, if young people with CFS/ME received adaptations and were still unable to attend school, they were questioned about the reason for their school absence, with the responsibility regarding school contact left to parents (Paper II). This fits with the previous finding, that adequate attitudes towards and educational management for young people with CFS/ME could depend on information about the impacts of CFS/ME from healthcare providers to school personnel (120). Conversely, ambivalent and unclear information about a CFS/ME diagnosis and the implications for school performance could negatively impact and possibly complicate the understanding of significant adults. This indicates that experienced healthcare providers should be responsible for providing information about CFS/ME to school personnel.

To reduce the burden on the student, teachers communicated with parents about the students' needs (Paper III). Young people with CFS/ME, however, felt alone in coping with impairments from CFS/ME and wanted someone to talk to about their educational frustrations (Paper II). This indicates that young people with CFS/ME might need problem-solving support from teachers to cope with their illness and disability and potentially decrease their emotional regulation (126). However, how this kind of support is received by young people with CFS/ME could also depend on their functioning and HRQoL. Increased contact between young people with CFS/ME and teachers was experienced with online schooling during the COVID-19 pandemic (Papers II & III). This is a new finding that might prove useful to improving the teacher–student relationship and support with problem-solving for young people with CFS/ME.

Young people with CFS/ME who struggled to receive adequate educational adaptations experienced that it did not help even when the healthcare providers communicated with school personnel (Paper II). Conversely, the teachers, counsellors and school nurses in this study experienced that healthcare providers ensured that adequate information about the management of CFS/ME and interdisciplinary collaboration on measures for young people with CFS/ME were valuable (Paper III). However, it was a challenge that healthcare providers could have various explanations on what causes CFS/ME (Paper III). This confirmed previous findings that the uncertainty surrounding the aetiology of CFS/ME makes it difficult to explain the disease to significant others, and thus, collaboration on the

management of adaptations and reintegration into school for young people with CFS/ME might be challenging (78). Second, it was also a challenge that interdisciplinary collaboration was introduced only after a diagnosis was set (Paper III). The challenges that school personnel experienced before the students were given a CFS/ME diagnosis represented a new and important finding in addressing the need for early interdisciplinary collaboration on measures for young people with CFS/ME symptoms.

The young participants with CFS/ME experienced that not all school nurses were knowledgeable about the impacts of CFS/ME (Paper II). The school nurse participants in this project (Paper III) were experienced with CFS/ME, and thus, their expertise in preventive healthcare made them think about possible preventive strategies for the young people with CFS/ME. This included early recognition of CFS/ME symptoms in young people and advising teachers about early educational adaptations (Paper III). This aligns well with previous suggestions that school nurses could play an important role in the early recognition of CFS/ME symptoms in young people (162) and that they can advise school personnel about the health and needs of affected students (231). However, a new finding was that school nurses might need to be educated about CFS/ME to be able to recognize CFS/ME symptoms and give advice on adequate adaptations.

To summarize, it was found that the teachers, counsellors and school nurses in this study were aware of the need for special attention towards educational and social challenges in young people with CFS/ME (Papers II & III). Nevertheless, the provision of adaptations was challenging for school personnel before they received information from healthcare providers (Paper III). Thus, young people with CFS/ME could be at risk of educational and social deficits from the time they started losing function until a diagnosis was given (Paper II). This might provide some explanation regarding reports by the young people with CFS/ME of severely impaired HRQoL (Paper I), which corroborated previous literature (127, 128, 130). The association between HRQoL and the management of educational and social adaptations at school for young people with CFS/ME is further discussed below.

6.2 Health-related quality of life and positive or negative contextual factors

The teachers, counsellors and especially school nurses were concerned about how the educational and social deficits impacted young people with CFS/ME (Paper III). These young people found it difficult to know their own limitations, and it could be excruciating for them to accept their impairments and that they could not develop in the same manner as their peers.

They could worry about school absence and become uncertain about their future (Paper II). In line with previous findings, the young people had challenges with symptoms and functioning (75) and, thus, might have experienced uncertainties and worry about their future (82, 102, 154). This indicates that these young people might need improved support with their health and challenges and advice regarding their future.

High levels of school absence mean that young people with CFS/ME were absent from the arenas where they would normally develop (Papers I & II), which was also challenging for the school personnel responsible for facilitating development (Paper III). In line with previous findings, young people with CFS/ME suffered from social isolation and impaired possibilities to develop among peers (82, 130). This may impair the development of competency in adolescent domains, including academic, cognitive and social skills (125). Conversely, young people with CFS/ME who had experienced adequate educational and social adaptations at school managed to participate in arenas where they could develop among peers (Paper II), and thus, this could have contributed to higher HRQoL scores (Paper I). This was in line with previous findings that HRQoL in young people with CFS/ME is associated with educational and social adaptations at school and positive or negative contextual factors (134). Thus, school personnel's suggestions of early interdisciplinary collaboration in managing of CFS/ME among young people (Paper III) might be important for the improvement of HRQoL in young people with CFS/ME. However, higher HRQoL scores and school contact could also have been due to factors like fewer health problems.

The severely impaired HRQoL found in young people with CFS/ME in this study (Paper I), confirmed that young people with CFS/ME suffer due to their illness being at a critical developmental period of life, namely, adolescence or young adulthood (100, 101). These young people did not like to think about the future (Paper II), which might be related to the age-dependent development of abstract thinking and difficulty imagining the long-term outcome of their disease (114). Rather, they focused on the present, and their attitudes towards their disease were to adapt, focus on what they were able to do and try to stay positive (Paper II). This aligns with previous findings that CFS/ME requires acceptance and taking a new situation into account to be able to go on with life (67, 103-105). The young people's developmental age and the severe impact of their disease indicate that they may benefit from adequate interdisciplinary management and support to improve HRQoL.

Interdisciplinary collaboration on educational and social adaptations for these young people are further discussed below.

6.3 Interdisciplinary collaboration on adaptations

The suggested strategies from school personnel and school nurses for preventing educational and social deficits in young people with CFS/ME (Paper III) are discussed in combination with previous recommendations for managing CFS/ME and finally, a new model for managing the disease in young people is suggested.

Young people with CFS/ME experienced that school personnel lacked knowledge about CFS/ME, and thus, adequate adaptations were not always available (Paper II). Information about adaptations for young people with CFS/ME from healthcare providers to school personnel was considered adequate and valuable but in need of earlier initiation (Paper III). The challenges experienced by the teachers, counsellors and school nurses in this study in the period before they were informed by healthcare providers made them suggest factors that could potentially improve management and prevent educational and social deficits in young people with CFS/ME (Paper III). The suggested factors were to problematize school absence early and to initiate interdisciplinary collaboration around early educational and social adaptations at school for young people with CFS/ME symptoms. To manage this, it was important not to wait until a diagnosis (Paper III). In line with previous recommendations, early initiation of measures for young people with CFS/ME is beneficial (3) and may contribute to the prediction of the long-term outcome of CFS/ME (135).

Adequate educational and social adaptations at school and the maintenance of contact with teachers and the school community were associated with higher levels of HRQoL in young people with CFS/ME (Papers I, II & III). These findings may indicate that early educational and social adaptations can be helpful for future functioning and HRQoL in young people with CFS/ME

The different solutions to educational and social challenges experienced by young people with CFS/ME (Paper II) were seemingly related to the challenges described by the teachers, counsellors and school nurses with experience in educational and social adaptations for young people with CFS/ME (Paper III). These findings indicate that the impact of a management plan for the education and social life of young people with CFS/ME may depend on the attitudes and actions of significant adults (232).

Suggestions from teachers, counsellors and school nurses on the early initiation of interdisciplinary collaboration and improvement measures, from the moment young people begin to lose function until a diagnosis is given might prevent some of the educational and social deficits experienced by young people with CFS/ME (Paper III). This is in line with the previous finding that early recognition of CFS/ME and persistent long-term interdisciplinary follow-up might reduce morbidity in young people with CFS/ME (119).

The school nurses' suggestions that they could play an important role by recognizing CFS/ME symptoms in students and subsequently advising teachers about early adaptations (Paper III) confirmed that they were a natural link between student, parents, teachers and healthcare providers (231). It also confirmed the school nurses' ability to initiate interdisciplinary collaboration around students with CFS/ME through referrals for assessment and initiation of advice on measures (162).

The role of teachers in CFS/ME follow-up has hitherto been poorly explored. Long-lasting school absence and lacking teacher–student relationships (Papers II & III) might cause teachers to lose confidence in their own capability to help affected students (161, 233). Thus, it might be particularly important to educate school personnel about impairments from CFS/ME and the subsequent need for adaptations in these young people's lives (3). This, to provide substantial support for young students with CFS/ME and their ability to go on with life, in line with needs described both for young people and adults with CFS/ME (232). The uncertainties that teachers experience regarding CFS/ME may engender reactions similar to those evoked when the teachers are responsible for students with cancer (234). These experiences might lead to mixed feelings about how to deal with the situation. Difficulty knowing how to help the affected student to remain in school and how to answer questions from other students about the disease while continuing to teach the rest of the class might be challenging for teachers (234). Teachers may also be concerned that the class setting might be affected when a student with CFS/ME is included in the mainstream class (167).

When interdisciplinary collaboration was initiated, healthcare providers communicated adequate and valuable information to school personnel about the impact of CFS/ME on young people (Paper III). This confirmed that specialized healthcare providers with specific competence in CFS/ME can inform school personnel about the impact of CFS/ME and adequate adaptations for young people with CFS/ME (3).

School personnel and school nurses also suggested that it was important to maintain teacher–student relationships and, thus, secure school contact for young people with CFS/ME (Paper III). In line with previous findings, this underscores the importance of school contact for young people's educational, social and emotional development (167), as well as guarding against feelings of being devalued (170).

A third suggestion from the school personnel and school nurses were to exchange competence between schools about adaptive measures for young people with CFS/ME and to hear from students' previous experiences with CFS/ME to increase knowledge among school personnel about CFS/ME and useful measures (Paper III). It has been found that adequate accommodations can be provided to improve academic skills in young people with CFS/ME (3, 235). Advice on how to teach young people with CFS/ME has also been provided by CFS/ME action groups (236). In Norway psychologist Ketil Jacobsen at St. Olavs hospital has been a pioneer in this, but this can be supplemented by exchange in competences between schools.

Previous recommendations on important factors for managing of CFS/ME in young people include a focus on symptoms and functional improvements through a flexible long-term management plan (3, 232, 237, 238); awareness of the normalizing role of school attendance and that home education lacks the important social and emotional development aspects found in peers (167) and, finally, mapping the illness beliefs about CFS/ME in parents and how they cope with the situation (237). It has previously been found that parents of young people with CFS/ME usually push their child to go to school at the beginning of the disease (11). They eventually gain awareness that PEM from physical and cognitive activity worsens symptoms, propelling them towards a protective position for their child (11, 237). Furthermore, some parents might find it difficult to understand why their child is unable to attend school, and they keep pushing them despite worsening symptoms (3, 237). Concern from parents, overprotection or difficulty understanding CFS/ME might be problematic for managing CFS/ME or re-entry into school for the young people involved (167). Thus, mapping of parents' illness beliefs and how they cope might be important.

Occupational therapists are usually engaged in identifying potential obstacles, solutions and provision of subsequent advice to educators to ensure individualized measures for young people with CFS/ME (239). A recent suggestion was that general practitioners should assess the educational challenges in young people with CFS/ME and provide early and detailed

information to schools about these young people's specific needs (120). Thus, interdisciplinary collaboration between specialist healthcare and school personnel to develop and distribute a manual for this to general practitioners, should be provided.

Since this project was initiated, new research has confirmed that HRQoL in young people with CFS/ME is lower than in healthy controls and young people with other chronic diseases, such as asthma, diabetes mellitus and cystic fibrosis (4). It has been confirmed that young people with CFS/ME struggle to different degrees and that symptoms and fluctuation of symptoms are individual in nature (11). Recent findings note that young people with CFS/ME have lower expectations about their physical exercise achievement and pre-exercise anxiety due to previous difficult experiences and that this might impact their future exercise efforts (240). Young people with CFS/ME have also demonstrated significantly higher rates of school absence, poorer school-related quality of life, poorer connectedness with school, and reduced academic performance compared to healthy controls (168). Cognitive behavioural treatment targeting fearful activity in young people with CFS/ME may improve activity in young people with severe fatigue (88). In addition, an online-based treatment programme for a six-month period has been deemed acceptable (241).

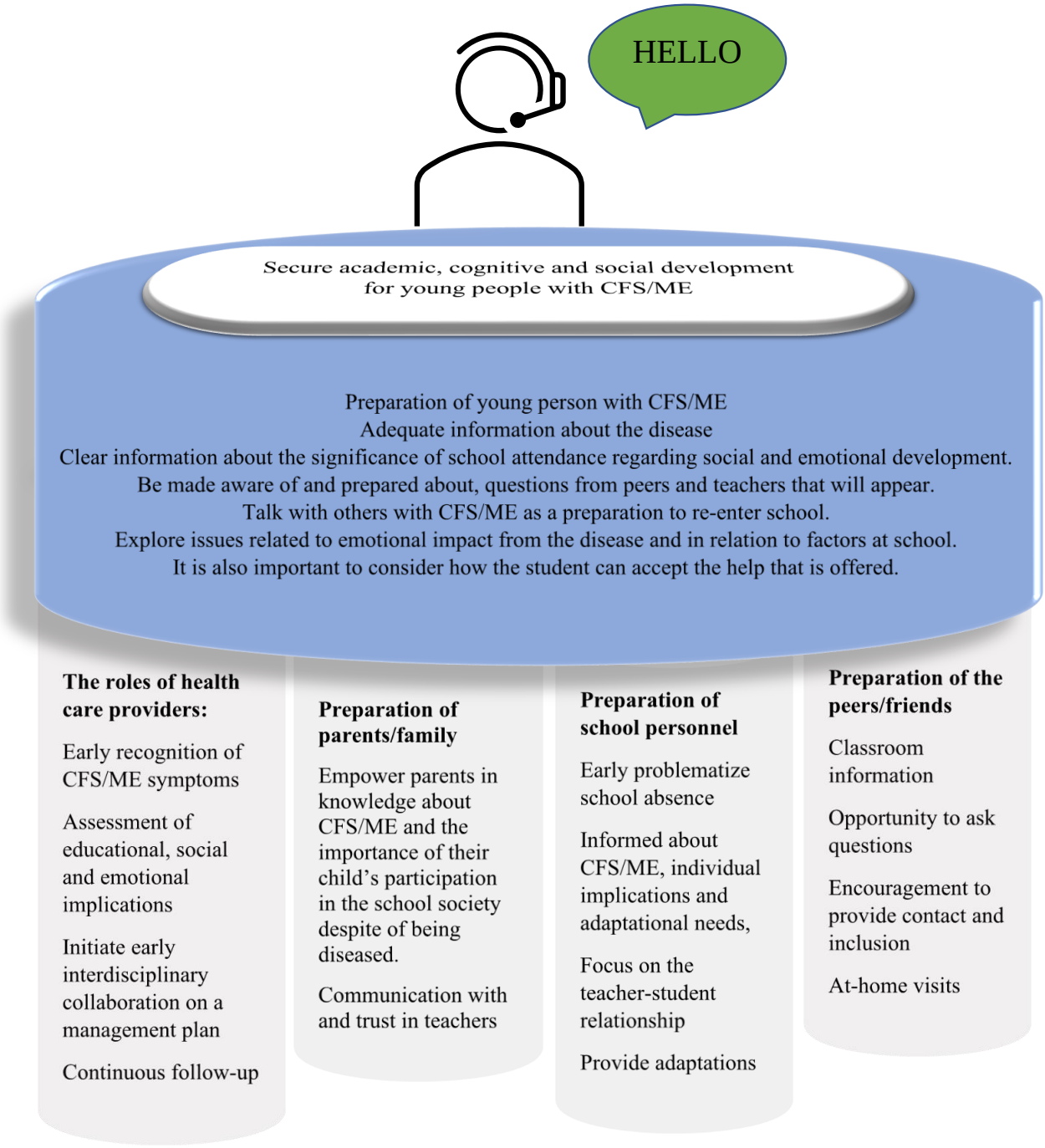
To understand the impact from CFS/ME on school functioning and inform targeted strategies for educational outcomes for young people with CFS/ME, it has recently been found that improved assessment characterizations of young people's school experiences are necessary (80). A new PROM especially for young people with CFS/ME is under development (135), and PEM, cognitive impairment and orthostatic intolerance have been suggested as core symptoms in paediatric CFS/ME (4). The European Network on ME/CFS (EUROMENE) has also published an expert consensus on the diagnosis, service provision and care for people with CFS/ME (242), and teachers' experiences with educational adaptations for children in primary school have recently been described for the first time (161).

Overall, the combined findings of this project and previous literature could form the basis for a new model for management of CFS/ME in young people as presented below. Young people with CFS/ME value help to move on with their lives (243). To rebuild hope for a better future, they might need support to accept and adjust their self-image from the impact of CFS/ME (67). For longterm functioning, it may be important that they remain engaged in education (12). Thus, the suggested model of this thesis might be helpful to improving HRQoL by improving the management of CFS/ME in young people with CFS/ME.

The findings of this study were combined with previous literature and illustrated in Figure 7 as a new model for interdisciplinary collaboration and knowledge exchange with concern to educational and social adaptations for young people with CFS/ME. Significant adults include healthcare providers, school personnel and parents. Peers are also significant participants in the management measures for securing academic, cognitive and social development among young people with CFS/ME. Individual factors for young people with a chronic disease for managing the education and social life at school include factors associated with the illness and treatment; variables relating to the young people such as individual response to the illness; academic impairment and social dysfunction; attitudinal issues of significant adults and the availability of educational and healthcare resources within the young people's school and healthcare systems (167).

6.4 A suggested new model for managing CFS/ME in young people

Figure 7. The HELLO platform for interdisciplinary exchange of competence on CFS/ME. **H**ealthcare providers, **E**ducators and **L**earning environments preventing **L**osses in young people with CFS/ME using interdisciplinary **O**nline devices.



7 Conclusion

This project confirmed that young people with CFS/ME have severely impaired HRQoL and it showed that there are some challenges related to educational and social adaptations at school that could positively or negatively impact the course of school participation. The challenges found in this project were related to uncertainty surrounding the CFS/ME diagnosis, difficulty maintaining teacher – student relationships and continuity of education and loss of confidence in school among young people with CFS/ME, especially in the period between the loss of function and the diagnosis. This substantiates the view that interdisciplinary collaboration around educational and social adaptations needs to be initiated earlier than is currently provided. In this project, suggestions for improving educational adaptations and social life at school also included a focus on teacher – student relationships, and exchange of experiences between schools. This could prevent losses and secure educational and social development in young people with CFS/ME.

These improvements require interdisciplinary collaboration around young people with CFS/ME before a diagnosis is set, that is, when the young people start losing function. This will require that schools problematize school absence early and that CFS/ME symptoms are recognized early. School nurses/school health services and general practitioners need to be educated to be able to recognize CFS/ME symptoms early in young people and, thus, initiate the necessary early interdisciplinary collaboration. These personnel should receive education on the implications of CFS/ME and what individual needs should be assessed for young people with CFS/ME, which should be informed in the context of interdisciplinary collaboration where a flexible plan for adaptations and follow-up is determined.

The current project had limited resources, and implementation for practice and research require new funding. With more funding, a prospective study exploring educational and social adaptations for young people with CFS/ME could provide information of relevance to improving both the management of CFS/ME and HRQoL in young people with CFS/ME.

8 Implications for Practice

It has previously been described that early management of CFS/ME can predict disease progression (66). Thus, early interdisciplinary collaboration on educational and social adaptations at school to prevent the deficits experienced by young people with CFS/ME may be important for the improvement of these young people's HRQoL.

Interventions aimed at improving HRQoL in young people with CFS/ME should focus on an early interdisciplinary management plan requiring that young people with CFS/ME symptoms are recognized when they start losing function. Problematizing school absences early on and referrals to school health services could be the first step. It has previously been found that school nurses could recognize CFS/ME in young people (236). In the current study, it was found that not all school nurses had knowledge about CFS/ME, and thus, they need to be educated to be able to recognize CFS/ME symptoms.

This study found that an early measure was to focus on the teacher – student relationship. Online education and communication were deemed to increase contact between teachers and students. Thus, online contact between the teacher and student should be continued for young people with CFS/ME when they are homebound. This may also improve reintegration into school.

To improve competence and attitudes towards CFS/ME in school personnel and healthcare providers and possibly maintain education for young people with CFS/ME, consistent, prospective identification and management of problems may be required (167). This might be possible if an interdisciplinary platform for collaboration and exchange of competence and experiences is developed especially to manage young people with CFS/ME.

Median time from functional impairments to CFS/ME diagnosis could be 15 months. An intervention to decrease this period may be to educate general practitioners about CFS/ME by connecting them to the suggested interdisciplinary platform for exchange of competence on CFS/ME, especially for young people. This may reduce the time from first contact with the general practitioner to the specialist referral. Interventions to prevent harm, that is, PEM during assessment should also be considered and highlighted in procedures in specialist healthcare services.

9 Implications for Research

This is the first study to explore factors associated with HRQoL in relation to educational and social adaptations for young people with CFS/ME. This is also the first study to explore how teachers, counsellors and school nurses experience educational and social adaptations at school for young people with CFS/ME. The study uncovered several areas of the study field that could be explored further.

For instance, future studies could examine how young people with CFS/ME prospectively experience the period from the loss of function to diagnosis, how they experience educational and social challenges, what adaptations are being offered to them and how this period specifically impacts their HRQoL, or how health problems impact school functioning. Second, it would be useful for the future management of CFS/ME to explore how young people with CFS/ME experience major transfers and, for advice on education, what young people value as important when they choose an educational track in high school. Third, exploring how online teaching impacts the school contact and functioning of students with CFS/ME could provide new and important knowledge for educational and social adaptations for these young people.

A further exploration of school personnel challenges with educational and social adaptations at school for students with CFS/ME could provide more nuances about how adaptations are managed and what information teachers lack to initiate early measures. Also important are the measures that teachers provide that work fine and why, that is, how digital schooling and contact enhance the teacher – student relationship.

It would also be important to explore how interdisciplinary collaboration meetings are carried out and how information and advice from healthcare providers to school personnel is transferred; thus, how they educate school personnel without transmitting uncertainty about CFS/ME could be important for improved knowledge exchange. Second, it would be important to explore healthcare providers' knowledge about or uncertainties regarding the aetiology of CFS/ME and how this impacts the information they exchange with schools. How healthcare providers perceive their engagement with the care for young people with CFS/ME could provide important information about the load on these providers. Third, it would be important to explore how school nurses recognize CFS/ME, the state of their resources to engage with this as well as their educational needs.

It is of most importance to explore how school absence can be problematized early for young people with CFS/ME symptoms and how school should act in order to refer students for further assessment in school health services without stigmatizing them. Finally, exploring how to reintegrate young people with CFS/ME into school, how health problems due to CFS/ME impact school maintenance and how school reintegration impacts educational, cognitive and social development and HRQoL in young people with CFS/ME is important.

Overall limitations to this study

This study included only a small sample of participants from Norway, and thus, the results might not be transferrable to all young people with CFS/ME or to school communities in any country. In this study exploration of young people with CFS/ME was not combined with their respective school personnel and school nurses, but with an independent sample of school personnel and school nurses. In future research this should be avoided, since a closer relationship between young people with CFS/ME and significant adults will probably provide better insight into the current setting under investigation.

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Papers I-III

Paper I

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Health and Quality of Life Outcomes

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Health-related quality of life in Norwegian adolescents living with chronic fatigue syndrome

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Abstract

Purpose: The primary aim was to measure health related quality of life (HRQoL) in a Norwegian cohort of adolescents with Chronic Fatigue Syndrome (CFS/ME). A secondary aim was to identify factors before diagnosis, at time of diagnosis and after diagnosis that were associated with HRQoL.

Methods: In this cross-sectional population-based study, HRQoL was measured by Pediatric Quality of Life Inventory™ Generic Core scale version 4.0 (PedsQL4.0) in 63 adolescents with CFS/ME. In addition, fatigue was measured by PedsQL Multidimensional Fatigue scale (PedsQL-MFS), depressive symptoms were measured by the Short Mood and Feelings Questionnaire (SMFQ), and disruption in school activities was measured by The De Paul Pediatric Health Questionnaire (DPHQ-N). Data were also collected from medical records and patient interviews.

Results: Age at diagnosis was 15 (2) years (mean (SD)), and four out of five participants were female. Time from diagnosis to reply was 39 (22) months. Adolescents with CFS/ME reported PedsQL4.0 score 50 (17), and boys reported a better score than girls (64 vs 47, 95% Confidence Interval (CI) for difference (− 27; − 6)). There were positive associations between overall HRQoL and support from a schoolteacher, school attendance or participation in leisure activities. There were negative associations between overall HRQoL and delayed school progression, having been to rehabilitation stay and depressive symptoms.

Conclusion: HRQoL in adolescents diagnosed with CFS/ME was low. The associations between reported HRQoL, healthcare previously provided, support from a schoolteacher, school attendance and participation in leisure activity may provide information of value when developing refined strategies for healthcare among adolescents with CFS/ME. Possible causal relationships must however be explored in future studies.

Keywords: Adolescent, Adolescent health, Chronic fatigue syndrome, Quality of life

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Introduction

Chronic fatigue syndrome (CFS) is characterized by overwhelming and severe disabling fatigue, and loss of physical and mental endurance [1]. The condition is also cited as Myalgic Encephalomyelitis (ME) to conceptualize a specific neuroimmunological condition [2]. A main characteristic of the disease is post exertional malaise (PEM). Other symptoms include orthostatic intolerance and other signs of autonomic dysfunctions, cognitive impairment, unrefreshing sleep, sore throat, headache, dizziness, heat and cold intolerance, muscular and abdominal pain, nausea, vomiting and mood disturbances [3–6]. CFS/ME occurs more frequently in the age groups 11–19 and 30–39 and is 3–4 times more common in girls than boys. In Norway 0,1–1,0% adolescents are affected [1, 7, 8]. Adolescents CFS/ME starts with an acute or gradual, infectious or non - infectious onset. Establishment of the diagnosis is frequently delayed, with the period from start of symptoms to diagnosis varying from 5 to 17 months [3, 9]. The CFS/ME disease course is often measured in years, and it is common to experience recurring improvement and relapses [1]. Among adolescents, CFS/ME is the most common cause of long-term absence from school [4, 10–13].

Previous studies have shown that CFS/ME severely impact health-related quality of life (HRQoL) [1]. Typically, adolescents with CFS/ME report lower HRQoL than adolescents diagnosed with other chronic health conditions like ADHD, cancer or cerebral palsy [14–17]. In an earlier Norwegian study of HRQoL among adolescents with CFS/ME, patients scored 49 whereas healthy controls scored 93 on a 0–100 generic HRQoL scale based on PedsQL 4.0 [16].

CFS/ME patients need care from primary health care and schools. Because of the complexity and severity of the illness, specialized care from personnel experienced with CFS/ME is often warranted, but frequently not available [1]. Despite substantial efforts from health care and schools, we still lack knowledge about effective strategies to improve disease outcome and HRQoL. The main aim of this study was to measure HRQoL in adolescents with CFS/ME, and a secondary aim was to identify factors before diagnosis, at diagnosis and after diagnosis positively or negatively associated with HRQoL.

Methods

Study design

A cross-sectional, population-based study of HRQoL in adolescents diagnosed with CFS/ME.

Study population

CFS/ME adolescent patients

Adolescents diagnosed with CFS/ME at St. Olavs or Oslo University Hospitals in Norway with age 12–18 at the time of diagnosis were invited by mail to participate. Participants were asked to complete a questionnaire and to attend an interview. Invitation was sent between August 2017 and January 2018, and time since diagnosis varied from 1 to 118 months. Of 168 invited, 86 (51,2%) agreed to participate, and 63 (37,5%) returned completed questionnaires. All participants were diagnosed with G 93.3 CFS/ME according to Jason diagnostic criteria [5], and the diagnosis was verified by an independent evaluation of medical records. Exclusion criteria were not being able to read Norwegian or reply to questionnaires or participate in interview. No one was excluded according to these criteria. Data collection from questionnaires, interviews and medical records finished in June 2018.

Measures

PedsQL generic Core scale

The Norwegian version of Pediatric Quality of Life Inventory™ Generic Core scale version 4.0 (PedsQL4.0) was used to measure HRQoL. PedsQL4.0 is a 23-item generic questionnaire developed to measure HRQoL in both healthy and acute or chronic ill children and adolescents [18]. A young adult version was used for ages 18–22. The PedsQL4.0 provides a generic sum score and subscale scores; Physical functioning (8 items) and Psychosocial functioning as total of: Emotional functioning (5 items), Social functioning (5 items) and School functioning (5 items). Participants were asked to rate each item during the last month on a Likert scale from 0 (never a problem) to 4 (almost always a problem). The items were reversely scored and linearly transformed on a scale ranging from 0 to 100 (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0). Higher scores indicate better HRQoL [18].

PedsQL multidimensional fatigue scale

The Pediatric Quality of Life Inventory™ Multidimensional Fatigue scale (PedsQL-MFS) was used to measure fatigue severity [19]. PedsQL-MFS is a generic scale with 18 items, and with subscale scores for three domains; general fatigue (6 items), fatigue related to sleep/rest (6 items) and cognitive fatigue (6 items). The response scale is the same as for PedsQL4.0. Higher scores indicate less fatigue.

Short mood and feelings questionnaire

The Short Mood and Feelings Questionnaire (SMFQ) is a self-report-form measuring depressive symptoms in children and adolescents. Thirteen items collectively describe depressive symptoms, covering for symptoms specific for major depression in DSM-IV. The SMFQ items have three reply options; “True” =2, “Sometimes” =1, “Not true” =0 referring to the last 2 weeks with a sum score from 0 to 26. A sum score of 11 or higher indicates depressive symptoms which possibly require treatment [20].

De Paul pediatric health questionnaire – Norwegian version

The De Paul Pediatric Health Questionnaire (DPHQ-N) for children and adolescents was translated to Norwegian by The Norwegian National Advisory Unit on CFS/ME. Translation and re-translation was performed according to EORTC [21], re-translated by one and accepted by original author. This is a self-report questionnaire for children ages 10–17 in three parts; 1) demographic data, 2) a list of CFS/ME-related symptoms from the current CFS/ME criteria according to Jason, with symptoms rated in frequency (0 = never, 1 = almost never, 2 = half the time, 3 = almost always, 4 = always), and in severity (0 = no problem, 1 = small problem, 2 = moderate problem, 3 = big problem, 4 = very big problem), and 3) experience of disruption in school activities or performance due to fatigue or cognitive difficulties [5, 22, 23].

Data from medical records and additional interviews with patients

Data from medical records were collected using a semi-structured guide developed by the research group. One pediatrician from each hospital and one psychiatrist from St. Olavs hospital used the same guide when collecting data from diagnostic evaluation in medical records. Supplementary data providing information of contact with primary health care personnel and schools were collected directly from the participants via a six to 7 minute telephone interview using an interview guide with the same questions for all participants.

Statistical analyses

First, we examined the study population characteristics. Then we examined HRQoL, fatigue and depressive symptoms in relation to study population characteristics. Continuous data (age, duration of fatigue, HRQoL, fatigue severity and depressive symptom scores) are presented as mean (SD), Median (Q1-Q3), and 95% confidence intervals (CI) where appropriate. Correlations between HRQoL, fatigue severity and depressive symptoms are presented as Pearson's correlation coefficients. Categorical data (gender, recovered/not recovered, delayed school progression and participation in leisure activities) are presented as numbers and percentages.

Next we examined a wide spectrum of factors related to the periods before diagnostic evaluation, through diagnostic evaluation and after diagnosis in relation to HRQoL (PedsQL 4.0). For these dichotomous factors two-sided independent sample t-test were used to assess differences. Complete results are presented in Supplemental Tables 1, 2, 3, 4, 5, 6, while statistically significant variables (5% level) are presented in Table 3. Generic HRQoL scores are presented as mean (SD) plus 95% confidence intervals (CI) for differences. For dimension scores only difference and CI are reported. These analyses were indicators for regression analysis and were not corrected for multiple testing.

Finally, we examined independent variables significantly associated with HRQoL as predictor variables in a multiple linear regression model, controlling for gender. The model included all participants with replies to dichotomous variables. Dependent variable was tested for normal distribution and outliers. Predictor variables were tested for multicollinearity. Unstandardized Beta coefficients with confidence intervals (CI), Adjusted R^2, model significance (ANOVA), F-values, β-values and *p*-values for each predictor were reported. A difference of 10 in primary outcome (HRQoL) was predetermined as clinically relevant.

Among 63 participants, 48 answered additional questions about contact with primary health care personnel and school. All statistical analyses were performed using SPSS version 23.

Ethics

The study was approved by The Regional Ethical Committee for medical and health profession research in Norway (REK 2017/749). All participants signed an informed consent, and the study was performed according to the declaration of Helsinki. The participants were offered a consultation to explore the need for further health care.

Results

Study population characteristics

Sixty-three adolescents with CFS/ME were included in the study, with a female: male ratio of 4,2:1 and mean age18 years (Table 1). Duration of fatigue initial to diagnostic evaluation was 15 months (10–33) (median (Q1-Q3)), and at the time of study enrollment 52 months (36–67) (median (Q1-Q3)). 37 (76%) of the adolescents had a delayed school progression defined as not having completed all compulsory subjects in school at their level. Furthermore, most of the adolescents (66%) reported no participation in leisure activities. Four adolescents had recovered after 6, 12 and 36 months, respectively (one unknown) (Table 2).

Generic PedsQL4.0 score for all participants in this study was 50 (17) (mean (SD)) (Table 1). The subscale scores were lowest for the domain school functioning and highest for social functioning. Overall PedsQL-MFS score was 36 (19). The domain general fatigue had the lowest score. There was a strong correlation between generic PedsQL4.0 and overall PedsQL-MFS scores (Pearson's $r = .861$, $p < .001$). The SMFQ sum score was 7 (5) (mean (SD)), whereas 27% of the participants scored 11 or higher, suggesting a possible treatment-requiring depression. The correlation between SMFQ

Table 1 Patient characteristics and overall results from PedsQL 4.0, PedsQL-MFS and SMFQ

	N	%	Mean (SD)	Median (Q1-Q3)
Gender (Female / Male / Undetermined)	50/12/1	79/19/2		
Age at time of enrolment	63		18 (2)	
Duration of fatigue before diagnostic evaluation (months) **a)**	48			15 (10–33)
Duration of fatigue at time of enrolment (months)	59			52 (36–67)
Recovered from CFS/ME (yes/no)	4/58	6/94		
Delayed school progression (yes/no)	37/12	76/24		
Participation in leisure activities (yes/no)	21/41	34/66		
PedsQL4.0				
Overall HRQoL score **(b)(c)**	63		50 (17)	
Sub-scale scores: Social functioning	63		67 (16)	
Emotional functioning	63		56 (20)	
Physical functioning	63		42 (25)	
School functioning	58		41 (21)	
Psychosocial functioning	63		55 (16)	
PedsQL-MFS				
Overall Fatigue score **(b)**	62		36 (19)	
Sub-scale scores: Cognitive fatigue	62		41 (25)	
Fatigue related to sleep/rest	62		36 (19)	
General fatigue	62		32 (23)	
SMFQ (13 items) Sum score **(c)**	63		7 (5)	
Score < 11 / 11 or higher	46/17	73/27		

a) Not available data from 15 participants, b) Pearson's correlation = .861, $p < .001$ between generic PedsQL4.0 and overall PedsQL-MFS, c) Pearson's correlation = −.544, $p < .001$between generic PedsQL 4.0 and SMFQ sum score

Table 2 PedsQL generic and multidimensional fatigue scales and SMFQ sum score related to patient characteristics

	PedsQL Generic scale (23 items)			*PedsQL Multidimensional Fatigue scale (18 items)*			*SMFQ sum score (13 items)*		
	N	Mean (SD)	95%CI for diff	***N***	Mean (SD)	95%CI for diff	N	Mean (SD)	95%CI for diff
Gender									
Girls	50	47 (16)	*(−27; −6)*	49	33 (16)	*(−30; −7)*	50	8 (5)	*(2–6)*
Boys	12	64 (20)		12	51 (24)		12	4 (3)	
Age:									
< 16	10	57 (17)	*(−4–20)*	10	46 (13)	*(−2; − 24)*	10	6 (3)	*(−4–0)*
16+	53	49 (17)		52	34 (20)		53	8 (6)	
Status at response time									
Not recovered from CFS/ME	58	48 (16)	*(19–51)*	57	33 (16)	*(−61; −28)*	58	8 (5)	*(0–10)*
Recovered from CFS/ME	4	83 (13)		4	78 (15)		4	3 (4)	
Is your school progression delayed									
Yes	37	49 (16)	*(−27; −4)*	37	34 (19)	*(−31; −6)*	37	8 (5)	*(−1–6)*
No	12	65 (18)		12	52 (21)		12	5 (6)	
School attendance before diagnosis									
< 50%	39	50 (18)	*(−25–2)*	38	36 (19)	*(−29–1)*	39	8 (5)	*(− 2–6)*
50% or more	9	61 (21)		9	50 (25)		9	6 (5)	
Do you participate in leisure activity									
Yes	21	59 (17)	*(4–22)*	21	46 (19)	*(5–24)*	21	6 (4)	*(−5–0)*
No	41	46 (16)		40	31 (18)		41	9 (5)	

Two-sided Independent Samples T-test. Difference in N is due to one undetermined which gender, and that participation with questionnaire was higher than participation in interview

and generic PedsQL4.0 score showed moderate to strong negative correlation ($r = -.544$, $p < .001$).

There was no correlation between duration of fatigue at time of study enrolment and overall score of HRQoL, fatigue level or depressive symptoms (Fig. 1). CFS/ME diagnosis for all participants were confirmed from medical records. In DPHQ-N question 59 IV "Worst symptom right now" there was an option to mark "I am not ill" – 4 participants marked this option at the time of reply. This was recorded as recovery from CFS/ME. Adolescents recovered from CFS/ME reported higher HRQoL than those who had not recovered (83 vs. 48, $p < .001$) (Table 2).

HRQoL, fatigue and depressive symptoms versus study population characteristics

There was a significant gender difference in generic PedsQL4.0 score where girls scored significantly lower than boys (47 vs 64, $p = .003$). In subscale scores, girls scored lower than boys for all dimensions (data not shown). There was also a significant gender difference in PedsQL-MFS score where girls scored lower than boys, and a similar result was found in the SMFQ score (Table 2).

School attendance, delayed school progression or participation in leisure activities were not statistically significant associated with SMFQ scores (Table 2). However, both generic PedsQL4.0 and overall PedsQL-MFS scores differed between adolescents having or not having participated in leisure activities or delayed school progression, with higher scores for the adolescents who were able to participate in leisure activities and adolescents with a normal school progression. School attendance before diagnostic evaluation showed a similar trend when setting the cut-off at 50% school attendance, although not attaining statistical significance.

HRQoL versus selected factors before diagnosis, by the time of diagnosis or after diagnosis

To further explore factors positively or negatively associated with HRQoL, 34 variables collected from patients and medical records were selected and divided into three groups; *before diagnosis, by the time of diagnosis and after diagnosis*. Factors significantly associated with generic PedsQL4.0 or subscale scores are shown in Table 3.

Looking at the possible association between HRQoL and factors being present before the CFS/ME diagnose, we found that school attendance < 50% or using medications were associated with lower HRQoL in PedsQL subscales, but not with generic PedsQL4.0.

All adolescents had a physician involved in diagnostic evaluation. Beyond that, there were differences regarding the type of health personnel involved. Our analyses show that when either an occupational therapist, a physical therapist or a clinical nutritionist were involved, this was positively associated with the PedsQL4.0 subscale emotional functioning, but not with other subscales or generic score.

For the period after diagnosis, we found four factors associated with HRQoL. Support from schoolteacher was positively associated with generic PedsQL 4.0 (55 vs 41, CI (0.08–29)), and with the subscale scores for emotional and psychosocial functioning. Participation in leisure activity was positively associated with generic PedsQL4.0 (59 vs 46, CI (4–22)) and with the subscale scores for physical, social, school and psychosocial functioning. Been to rehabilitation stay was

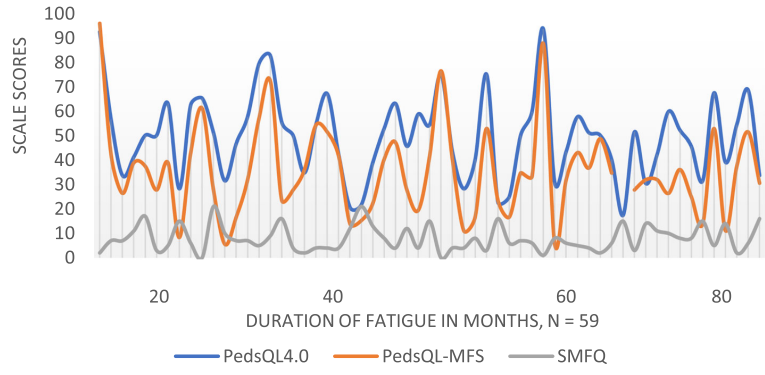

Fig. 1 HRQoL-, Fatigue- and SMFQ- scores in relation to Duration of fatigue, $n = 59$. For PedsQL4.0 and PedsQL-MFS high score is better, for SMFQ low score is better

Table 3 Selected factors present before, during or after diagnosis related to HRQoL as determined by PedsQL generic scale score and subscale scores

	N	Factors present							
		Overall Yes No HRQoL			Physical functioning	Emotional functioning	Social functioning	School functioning	Psychosocial functioning
	(Yes/No)	Mean (SD)	Mean (SD)	*Diff (CI)*	*Diff (CI)*	*Diff (CI)*	*Diff (CI)*	*Diff (CI)*	*Diff (CI)*
Factors before diagnosis									
School attendance before diagnosis < 50%	39/9	50 (18)	61 (21)	Ns	Ns	Ns	Ns	22 *(−39; −6)*	13 *(−26; −1)*
Using medications before diagnosis	17/30	45 (20)	56 (17)	Ns	Ns	Ns	13 *(−22; −3)*	Ns	10 *(−20; −1)*
Factors during diagnostic evaluation									
Occupational therapist engaged in diagnostic evaluation	31/18	53 (18)	49 (20)	Ns	Ns	−12 *(2–24)*	Ns	Ns	Ns
Physical therapist engaged in diagnostic evaluation	34/15	54 (17)	47 (22)	Ns	Ns	−14 *(3–26)*	Ns	Ns	Ns
Nutritionist engaged in diagnostic evaluation	5/44	61 (8)	51 (19)	Ns	Ns	−19 *(1–37)*	Ns	Ns	Ns
Factors after diagnosis									
Support from schoolteacher	42/7	55 (17)	41 (17)	−14 *(1–29)*	Ns	−19 *(4–35)*	Ns	Ns	−15 *(2–28)*
Been to physical therapy	27/22	50 (17)	57 (19)	Ns	Ns	Ns	Ns	14 (−27; −1)	Ns
Been to rehabilitation stay	14/35	43 (15)	57 (17)	14 *(−24; −3)*	Ns	23 *(−34; −11)*	14 *(− 22; −6)*	Ns	14 *(− 24; −5)*
Participation in leisure activity	21/41	59 (17)	46 (16)	−13 *(4–22)*	−18 *(6–31)*	Ns	− 13 *(5–21)*	−15 *(4–26)*	−10 *(2–19)*
Delayed school progression	37/12	49 (16)	65 (18)	16 *(− 27; −4)*	18 *(− 35; − 2)*	Ns	14 *(−23; − 6)*	21 *(− 35; −7)*	14 *(− 24; − 4)*
SMFQ Sum score 11 or higher (cut of, depression symptoms)	17/46	37 (12)	55 (16)	18 *(− 27; − 10)*	14 *(− 28; − 1)*	28 *(− 37; − 20)*	21 *(−28; − 14)*	13 *(−25; − 1)*	21 *(−29; − 14)*

Two-sided Independent Samples T-test. Overall HRQoL reported as mean (SD), and differences in overall HRQoL and subscale scores reported with 95% Confidence Interval or Ns if not statistically significant

negatively associated with generic PedsQL4.0 (43 vs 57, CI (– 24; – 3)) and with subscale scores for emotional, social and psychosocial functioning. Been to physical therapy was negatively associated with school functioning. Delayed school progression was negatively associated with generic PedsQL4.0 (49 vs 65, CI (– 27; – 4)) and with subscale scores for physical, social, school and psychosocial functioning. We also found that possible clinically significant depression (SMFQ score equal to or greater than 11) was negatively associated with generic HRQoL (CI (– 27; – 10)) and with all dimensions. Findings from bivariate analyses along with clinical relevance were indicators for which factors to include in the multiple regression analysis.

Multivariate analysis: HRQoL versus selected factors in a regression model

Multiple linear regression analysis was performed to predict HRQoL based on the four variables from after diagnosis, identified from bivariate analyses and with the most significant positive or negative association. The four variables were considered as clinically relevant and adequate to both health care, support from school and to the loss of important activities these adolescents suffer from. Dependent variable generic PedsQL 4.0 was normally distributed. Forthy-eight participants had responded to all predictor variables. Predictor variables correlated with HRQoL (Pearson's $r > .300$ except for support from teacher r .290) (Table 4).

The results from the multiple regression analysis confirmed the associations from bivariate analyses. The regression model was significant at the level $p = .001$, explaining 32% of the variance. Support from schoolteacher and participation in leisure activities were positively associated with HRQoL, while negative associations came from delayed school progression, and having been to a rehabilitation stay. The regression coefficient for delayed school progression was – 10 (β – .249), and for support from schoolteacher 10 (β .200), indicating clinical relevance. We also looked at the multiple linear regression analysis without the four participants who reported recovery. The predictors were distributed similarly in the regression model, and with similar results.

Based on the difference in bivariate analyses between participants with and without depressive symptoms, we looked at the multiple regression analysis if excluding the group of 10 participants with depressive symptoms. With $n = 38$ participants the distribution of the predictors was still the same, but with regression coefficient for delayed school progression at – 14 (β – .378), and support from schoolteacher 10 (β .162). Participation in leisure activity, β .260, and been to rehabilitation stay, β – .143. Hence the model explained 23% of the variance ($p = .019$).

Discussion

The primary aim of this study was to measure HRQoL in adolescents living with CFS/ME. Overall, HRQoL in this patient group was low. Secondary aims were to identify factors before, at time of and after diagnosis, associated with HRQoL. School absence higher than 50% before diagnostic evaluation, delayed school progression, having attended physical therapy or rehabilitation stay after diagnosis were associated with lower HRQoL. Occupational therapist, physical therapist or clinical nutritionist engaged in diagnostic evaluation were associated with higher HRQoL. After diagnosis, being supported by a schoolteacher, attending school or participating in leisure activities were associated with higher HRQoL. We found no correlation between duration of fatigue at time of enrolment and HRQoL, fatigue severity or depressive symptoms.

HRQoL in adolescents living with CFS/ME was low compared to healthy adolescents as reported by previous studies. Healthy adolescents typically scored 83 or higher, and adolescents with other chronic diseases scored from 66 to 77 [14–17]. Importantly, girls

Table 4 Multiple Linear regression - predictors to HRQoL in adolescents diagnosed with CFS/ME

$N = 48$	Beta coefficient	(95%CI)	β	p
Constant	50	(34–66)		.000
Gender (a)	10	(−1–21)	.230	.079
Support from schoolteacher (b)(c)	10	(−3–23)	.200	.121
Delayed school progression (d)	−10	(−21–1)	−.249	.051
Participation in leisure activity (e)	8	(−2–18)	.211	.114
Been to rehabilitation stay (f)	−8	(− 18–2)	−.212	.104

a) female =0, male =1), **b)** support from schoolteacher no =0, yes =1, **c)** support from schoolteacher, correlation coefficient to HRQoL .290 (<.3). **d)** school delay no = 0, yes =, 1, **e)** participate in leisure activity no = 0, yes =1, **f)** been to rehabilitation stay no = 0, yes =1
Model summary: Adj. R^2 .319, F Change 5.399, Sig (ANOVA) p = .001

scored lower than boys in both generic and dimensional PedsQL4.0 and PedsQL-MFS. Similar results were earlier found in a Norwegian study from 2015 [16], and internationally [24]. The low HRQoL scores in CFS/ME adolescents suggest a need for new strategies to improve HRQoL.

School absence higher than 50% before diagnostic evaluation or delayed school progression were associated with lower HRQoL. Maintaining contact with school have in previous studies shown to be important [25, 26]. CFS/ME symptoms and the subsequent reduction in activities, socializing and school delay may lead to anxiety, depressive mood and increased tension [27]. Measures to maintain school progression to improve HRQoL for adolescents with CFS/ME should be considered.

Participation in physical therapy or a rehabilitation stay were associated with lower HRQoL. Rehabilitation programs with exercise, mobilization and body awareness typically delivered from physical therapists, are earlier described as effective in reducing medium- and long-term fatigue severity in CFS/ME patients [28]. These findings seemingly conflict with our findings, i.e. that physical therapy or rehabilitation stay were associated with lower HRQoL. The lack of knowledge and disagreement about strategies to improve HRQoL in these patients might contribute to disruption in therapeutic alliances with patients and parents, and to distrust in health care personnel [29–31]. Our findings might indicate that adolescents with low HRQoL are more likely to attend or being offered a rehabilitation stay, and that a long-term plan with regularly mapping of symptoms could be most helpful for health care personnel to plan an individualized rehabilitation stay.

On the other hand, occupational therapist, physical therapist and clinical nutritionist engaged in diagnostic evaluation were associated with higher HRQoL. Occupational and physical therapists are commonly engaged in adaption of management plans after CFS/ME diagnosis [1]. Our findings suggest engagement also in diagnostic evaluation. Engagement from nutritionists could potentially be important since approximately 10% of adolescents with CFS/ME suffer from eating-difficulties [13].

Importantly, we found that support from schoolteacher was associated with higher HRQoL. To meet responsive and caring teachers, get assistance from sympathetic counselors, and the possibility to have flexible schedules might be just as important as support and care from health care professionals [1]. The positive association between support from schoolteachers and emotional functioning may be related to prevention of depressive symptoms. The importance of meeting in small groups with peers, and cooperation between health care professionals and school is earlier described as helpful [27]. The ability of schoolteachers to support adolescents with CFS/ME who are not present at school might improve the cohesion with school society and secondary improve HRQoL.

School attendance could result in PEM from physical, cognitive and social activity. Socialization is recommended as a priority when adolescents with CFS/ME attend school, and adjustment of the curriculum is necessary [1]. Teaching via digital tools is a strategy that potentially benefit adolescents with CFS/ME.

Participation in leisure activities was associated with higher HRQoL. Perhaps this is because the healthiest adolescents are more likely to participate in leisure activities. However, participation in leisure activities should be further studied throughout the course of CFS/ME when searching for strategies to improve HRQoL. This may decrease stigmatization of adolescents with CFS/ME who participate in leisure activities, even if they don't manage obligatory school activity in line with healthy adolescents.

We found no correlation between duration of fatigue at time of enrolment and HRQoL, fatigue severity or depressive symptoms. Adolescents with CFS/ME are not able to do the things they want, and they suffer from loss, disruption and coping barriers [1, 26, 27, 32]. A previous study found no statistical evidence between depressive symptoms and low HRQoL [16]. Rather the duration of fatigue before diagnosis, the demanding diagnostic process, lack of medical understanding and lack of positive prognosis information, might provoke anxiety since the adolescents perceive their CFS/ME as being permanent and threatening to their future hopes and dreams [6, 10]. Previous studies emphasize how important it is that professionals involved in the diagnostic evaluation and health care of adolescents after diagnosis of CFS/ME agree about treatment and communicate consistently [29]. According to Rowe [1] "Management of CFS/ME requires careful attention.", and that the surroundings are aware and supportive in order to give the adolescents a potential to prevent depressive symptoms and gain hope of an active and productive future. To avoid increased symptoms and relapses, they need a long-term plan for health care with regularly mapping of symptoms, guidance on activity, and regularly adjustments to symptoms severity or improvement [1]. Collectively, our findings support this.

Strengths and limitations

A strength to our study is that we explore the relationship between HRQoL and various factors especially during the period after diagnosis. Furthermore, our participants had a mean duration of fatigue close to 4 years which adds relevance to our findings. The diagnosis was verified according to the Jason criteria, and the diagnostic evaluation and health care after diagnosis at the two hospitals participating in this study are relatively uniform. A limitation to our study was a possibility to miss significant associations between HRQoL and factors before, by the time of or after diagnosis due to a small sample size. In the multiple regression analysis we only adjusted for gender, due to sample size. Bivariate analyzes were not corrected for multiple testing, and accordingly results should be cautiously interpreted. Sixty-three participants reported HRQoL with self-completed questionnaires, while only 48 responded to the telephone interview. This lost-to-follow-up bias resulted in exclusion of participants in the bivariate and multivariate analyses and could have been avoided by adding the questions from the telephone interview to the self-complete questionnaire. A further limitation was that patients with CFS/ME often have reduced cognitive function, and it may be difficult to remember exactly what occurred early in the disease course leading to recall-bias. Importantly, given our cross-sectional study design, it is impossible to conclude whether factors have a causal significance to HRQoL.

Implications

We still lack effective treatment of fatigue in CFS/ME patients, and despite effort from health-services and schools, HRQoL in adolescents with CFS/ME is low. A focus on strategies to improve psychosocial function, especially in relation to school, during diagnostic evaluation and after diagnosis might contribute to higher HRQoL (Fig. 2). Focus on participation in leisure activities in association to HRQoL is potentially needed to improve HRQoL and avoid stigmatization of CFS/ME adolescents. A long-term plan for health care with regularly mapping of symptoms from early stages of the disease might reveal data with importance to prevent the significant reduction of HRQoL, regarding both physical, social and emotional aspects [1]. Cooperation between schools, primary health care and hospitals when caring for adolescents with CFS/ME are of most importance.

For all patients with chronic health conditions, the goal of health care should be to restore them to the fullest health possible by improving symptom management, treatment adherence, and their ability to cope with their condition. For this reason, HRQoL may be just as important as biomedical measures when assessing patients with chronic health conditions like CFS/ME. The PedsQL4.0 is developed from the Worlds Health Organization's definition of health and is a valid instrument for this purpose [33].

Conclusion

In this cross-sectional study of adolescents with CFS/ME we found low HRQoL. The study identified new and possibly important factors associated with HRQoL. When exploring factors before, at the time of or after diagnosis associated with HRQoL, we found that school attendance, support from a schoolteacher and participation in leisure activities were associated with higher HRQoL. We also found associations to higher emotional functioning, when occupational therapist, physical therapist and clinical nutritionist were engaged in diagnostic evaluation. On the other hand, school absence higher than 50% before diagnostic evaluation, delayed school progression or having been to a rehabilitation stay were negatively associated with HRQoL. Early diagnosis, mapping of symptoms severity and HRQoL, maintaining school contact and early action to prevent depressive symptoms might be important to improve HRQoL in these patients. Limitations to our study design imply that future interventional studies are needed to confirm whether the identified factors can be used to improve HRQoL in adolescents with CFS/ME.

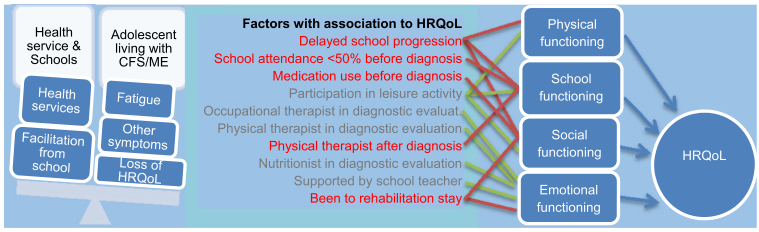

Fig. 2 Factors found with association to HRQoL in adolescents living with CFS/ME

Supplementary information

Supplementary information accompanies this paper at https://doi.org/10.1186/s12955-020-01430-z.

Additional file 1: Supplemental Table 1. HRQoL as measured by generic PedsQL4.0 versus selected factors before, at or after diagnosis **Supplemental Table 2.** Physical functioning versus selected factors before, at or after diagnosis. **Supplemental Table 3.** Emotional functioning versus selcted factors before, at or after diagnosis. **Supplemental Table 4.** Social functioning versus selected factors before, at or after diagnosis. **Supplemental Table 5.** School functioning versus selected factors before, at or after diagnosis. **Supplemental Table 6.** Psychosocial functioning versus selected factors before, at or after diagnosis.

Abbreviations

CFS/ME: Chronic fatigue syndrome/myalgic encephalomyelitis; PEM: Post exertional malaise; HRQoL: Health related quality of life; PedsQL4.0: Pediatric Quality of Life Inventory™ Generic Core scale version 4.0; PedsQL-MFS: Pediatric Quality of Life Inventory™ Multidimensional Fatigue scale; SMFQ: Short Mood and Feelings Questionnaire; DPHQ-N: De Paul Pediatric Health Questionnaire –Norwegian version

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Authors' contributions

WAS collected, analyzed and interpreted the patient data. TBR supervised the project, analyses and interpretation of data. VH supervised the project, analyses and interpretation of data. IBH facilitated and supervised data collection from Oslo University Hospital. CS supervised the project and collected data from medical records. HE collected data from medical records. All authors read and approved the final manuscript.

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Availability of data and materials

The dataset generated and analyzed during the current study is not public available due to ethical standards for treatment of patient data.

Ethics approval and consent to participate

All procedures performed were in accordance with the ethical standards of the national research committee (The Regional Ethical Committee for medical and health profession research in Norway, REK 2017/749). Informed consent was obtained from all individual participants included in the study.

Consent for publication

Informed consent obtained from all individual participants included consent for publication of anonymized data.

Competing interests

The authors declare that there is no competing interest.

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Functional Assessment Chronic Illness Therapy (Fatigue) (FACIT-F), Multi-Dimensional Assessment of Fatigue (MAF), Multi-Dimensional Fatigue Inventory (MFI), Pediatric Quality Of Life (PedsQL) Multi-Dimensional Fatigue Scale, Profile of Fatigue (ProF), Short Form 36 Vitality Subscale (SF-36 VT), and Visual Analog Scales (VAS). Arthritis Care Res (Hoboken). 2011;63(Suppl 11):S263–86.
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Supplemental Table 1: HRQoL as measured by generic PedsQL4.0, variables from questionnaires and data collected from patient journals.

Generic PedsQL4.0	N Yes/No	Mean (SD) Yes	Mean (SD) No	*p*
Factors before diagnosis				
Using medications before diagnosis	17/30	45.3 ± 19.8	55.8 ± 17.4	*.063*
School attendance before diagnosis < 50 %	39/9	49.7 ± 17.9	61.0 ± 21.0	*.104*
Factors at diagnostic evaluation				
Diagnosed in outpatient clinic (not hospitalized)	38/9	52.5 ± 16.9	53.5 ± 25.1	*.892*
Joint consultation by medical and psychiatric units	22/20	51.2 ± 18.5	53.0 ± 16.5	*.730*
Child/adolescent psychiatry engaged in diagn. eval.	45/4	52.0 ± 18.8	49.5 ± 19.0	*.797*
Nurse engaged in diagnostic evaluation	14/35	48.6 ± 17.9	53.0 ± 19.0	*.457*
Nutritionist engaged in diagnostic evaluation	5/44	60.7 ± 8.1	50.8 ± 19.2	*.264*
Physical therapist engaged in diagnostic evaluation	34/15	53.7 ± 17.0	47.4 ± 21.7	*.273*
Occupational therapist engaged in diagn. evaluation	31/18	53.2 ± 17.6	49.4 ± 20.4	*.495*
Educator engaged in diagnostic evaluation	7/42	63.3 ± 16.3	49.9 ± 18.4	*.076*
Social worker engaged in diagnostic evaluation	2/47	48.9 ± 20.0	51.9 ± 18.7	*.826*
Collaborative meeting with primary health care held	9/9	48.3 ± 27.6	58.1 ± 17.1	*.378*
Factors after diagnosis				
Individualized school schedule	30/19	51.6 ± 16.3	55.2 ± 20.3	*.495*
Individualized examination plan	17/32	49.5 ± 15.7	54.9 ± 18.9	*.327*
Home education by teacher	10/39	49.6 ± 22.6	53.9 ± 16.7	*.498*
Organized schooldays (with or without individualized school schedule)	43/6	52.0 ± 17.9	60.3 ± 16.8	*.289*
Support from schoolteacher	42/7	55.1 ± 17.4	40.8 ± 16.7	*.049*
Support from advisor in school	38/6	52.2 ± 18.0	47.6 ± 16.9	*.566*
Been seeing pedagogic-psychologic services	27/21	51.1 ± 18.5	56.3 ± 16.9	*.321*
Been seeing social worker	7/41	47.5 ± 16.6	53.0 ± 17.3	*.439*
Been seeing nutritionist	8/39	47.0 ± 12.7	54.0 ± 19.0	*.329*
Been seeing a psychiatric nurse or nurse	2/45	58.7 ± 12.3	51.8 ± 17.5	*.586*
Been seeing a school nurse	12/37	49.7 ± 14.4	54.1 ± 18.9	*.469*
Been to psycho motoric physical therapist	4/32	60.9 ± 14.7	52.3 ± 18.5	*.381*
Been to physical therapist	27/22	49.9 ± 16.6	56.8 ± 18.9	*.178*
Been seeing an occupational therapist	25/23	51.5 ± 17.8	53.0 ± 16.7	*.759*
Been to child and adolescent psychiatry	28/21	51.4 ± 17.8	55.2 ± 18.1	*.472*
Been seeing a general practitioner	37/11	52.0 ± 18.3	57.1 ± 17.4	*.416*
Responsibility group	19/30	55.7 ± 19.4	51.3 ± 16.9	*.401*
Been to rehabilitation stay	14/35	43.1 ± 15.2	57.0 ± 17.5	*.012*

Participated in disease-specific coping courses	18/31	52.0 ± 18.6	53.6 ±17.7	*.770*
Received aids (i.e. wheelchair, taxi-drive to school)	17/32	49.3 ± 15.6	55.0 ± 18.9	*.292*
Delayed school progression	37/12	49.2 ± 16.3	64.7 ± 17.9	*.007*
Participate in leisure activity	21/41	59.4 ± 16.5	46.0 ± 16.5	*.004*

Supplemental Table 2: HRQoL as measured by PedsQL4.0– physical functioning dimension, variables from questionnaires and data collected from patient journals.

PedsQL4.0 dimension- physical functioning	**N**	**Mean** (SD)	**Mean** (SD)	
	Yes/No	**Yes**	**No**	*p*
Factors before diagnosis				
Using medications before diagnosis	17/30	36.8 ± 24.4	47.6 ± 26.2	*.167*
School attendance before diagnosis < 50 %	39/9	41.7 ± 25.7	49.1 ± 28.8	*.447*
Factors at diagnostic evaluation				
Diagnosed in outpatient clinic (not hospitalized)	38/9	44.3 ± 24.0	42.4 ± 34.3	*.849*
Joint consultation by medical and psychiatric units	22/20	39.4 ± 26.3	45.9 ± 25.6	*.420*
Child/adolescent psychiatry engaged in diagn. eval.	45/4	43.2 ± 25.9	38.3 ± 29.5	*.723*
Nurse engaged in diagnostic evaluation	14/35	38.0 ± 24.1	44.7 ± 26.7	*.417*
Nutritionist engaged in diagnostic evaluation	5/44	45.9 ± 14.2	42.4 ± 27.0	*.779*
Physical therapist engaged in diagnostic evaluation	34/15	43.1 ± 26.3	41.9 ± 25.9	*.876*
Occupational therapist engaged in diagn. evaluation	31/18	42.7 ± 27.3	42.8 ± 24.2	*.996*
Educator engaged in diagnostic evaluation	7/42	59.7 ± 21.4	39.9 ± 25.8	*.061*
Social worker engaged in diagnostic evaluation	2/47	25.0 ± 30.9	43.5 ± 25.8	*.328*
Collaborative meeting with primary health care held	9/9	43.7 ± 37.0	48.8 ± 27.7	*.744*
Factors after diagnosis				
Individualized school schedule	30/19	44.1 ± 23.4	45.6 ± 28.9	*.848*
Individualized examination plan	17/32	40.8 ± 25.0	46.7 ± 25.8	*.444*
Home education by teacher	10/39	40.5 ± 33.8	45.7 ± 23.2	*.570*
Organized schooldays (with or without individualized school schedule)	43/6	43.4 ± 25.3	53.6 ± 27.1	*.361*
Support from schoolteacher	42/7	46.6 ± 25.6	33.2 ± 22.4	*.199*
Support from advisor in school	38/6	41.0 ± 24.5	49.5 ± 23.7	*.433*
Been seeing pedagogic-psychologic services	27/21	42.2 ± 27.5	48.5 ± 23.2	*.407*
Been seeing social worker	7/41	32.6 ± 27.9	45.6 ± 23.9	*.202*
Been seeing nutritionist	8/39	35.2 ± 19.1	46.0 ± 26.8	*.283*
Been seeing a psychiatric nurse or nurse	2/45	57.8 ± 2.2	43.0 ± 25.3	*.001*
Been seeing a school nurse	12/37	38.3 ± 24.6	46.7 ± 25.6	*.321*
Been to psycho motoric physical therapist	4/32	65.6 ± 23.2	42.8 ± 25.4	*.097*

Been to physical therapist	27/22	39.7 ± 25.2	50.8 ± 24.9	*.132*
Been seeing an occupational therapist	25/23	41.0 ± 26.4	46.6 ± 22.9	*.441*
Been to child and adolescent psychiatry	28/21	43.9 ± 24.6	45.7 ± 27.0	*.804*
Been seeing a general practitioner	37/11	44.1 ± 27.1	47.9 ± 20.4	*.665*
Responsibility group	19/30	52.3 ± 26.5	39.9 ± 23.9	*.097*
Been to rehabilitation stay	14/35	35.9 ± 16.5	48,2 ± 27,6	*.064*
Participated in disease-specific coping courses	18/31	45.2 ± 27.8	44.4 ± 24.4	*.915*
Received aids (i.e. wheelchair, taxi-drive to school)	17/32	39.7 ± 26.9	47.3 ± 24.6	*.324*
Delayed school progression	12/37	40.2 ± 24.9	58.5 ± 22.7	*.028*
Participate in leisure activity	41/21	54.6 ± 24.8	36.2 ± 22.2	*.004*

Supplemental Table 3: HRQoL as measured by PedsQL4.0 – emotional functioning dimension, variables from questionnaires and data collected from patient journals.

PedsQL4.0 dimension-Emotional functioning	**N**	**Mean** (SD)	**Mean** (SD)	
	Yes/No	**Yes**	**No**	*p*
Factors before diagnosis				
Using medications before diagnosis	17/30	51.2 ± 22.8	59.5 ± 17.4	*.167*
School attendance before diagnosis < 50 %	39/9	55.5 ± 18.2	62.8 ± 25.8	*.324*
Factors at diagnostic evaluation				
Diagnosed in outpatient clinic (not hospitalized)	38/9	57.2 ± 18.6	60.0 ± 25.1	*.709*
Joint consultation by medical and psychiatric units	22/20	60.0 ± 21.4	56.8 ± 15.1	*.576*
Child/adolescent psychiatry engaged in diagn. eval.	45/4	57.8 ± 20.4	51.3 ± 8.5	*.531*
Nurse engaged in diagnostic evaluation	14/35	58.9 ± 22.0	56.6 ± 19.0	*.709*
Nutritionist engaged in diagnostic evaluation	5/44	74.0 ± 7.4	55.3 ± 19.8	*.043*
Physical therapist engaged in diagnostic evaluation	34/15	61.8 ± 16.7	47.0 ± 22.5	*.014*
Occupational therapist engaged in diagn. evaluation	31/18	61.9 ± 17.4	49.2 ± 21.3	*.027*
Educator engaged in diagnostic evaluation	7/42	64.3 ± 16.7	56.1 ± 20.1	*.312*
Social worker engaged in diagnostic evaluation	2/47	77.5 ± 10.6	56.4 ± 19.6	*.139*
Collaborative meeting with primary health care held	9/9	49.4 ± 26.6	69.4 ± 11.8	*.056*
Factors after diagnosis				
Individualized school schedule	30/19	58.0 ± 18.8	60.0 ± 22.8	*.740*
Individualized examination plan	17/32	57.6 ± 17.5	59.4 ± 21.8	*.779*
Home education by teacher	10/39	59.0 ± 21.3	58.7 ± 20.2	*.969*
Organized schooldays (with or without individualized school schedule)	43/6	58.7 ± 20.4	59.2 ± 20.6	*.960*
Support from schoolteacher	42/7	61.5 ± 19.1	42.1 ± 20.2	*.017*
Support from advisor in school	38/6	60.4 ± 21.2	45.0 ± 15.8	*.097*

Been seeing pedagogic-psychologic services	27/21	60.0 ± 17.7	58.8 ± 22.7	*.839*
Been seeing social worker	7/41	56.4 ±14.9	58.3 ± 20.6	*.820*
Been seeing nutritionist	8/39	58.8 ± 19.4	58.7 ± 21.1	*.997*
Been seeing a psychiatric nurse or nurse	2/45	55.0 ± 28.3	57.7 ± 19.6	*.854*
Been seeing a school nurse	12/37	61.3 ± 17.9	58.0 ± 21.1	*.631*
Been to psycho motoric physical therapist	4/32	46.3 ± 19.3	58.9 ± 19.8	*.235*
Been to physical therapist	27/22	60.2 ± 20.1	57.0 ± 20.7	*.594*
Been seeing an occupational therapist	25/23	61.2 ± 17.2	54.6 ± 22.0	*.249*
Been to child and adolescent psychiatry	28/21	54.8 ± 21.8	64.1 ± 17.0	*.115*
Been seeing a general practitioner	37/11	56.9 ± 20.0	64.5 ± 21.7	*.280*
Responsibility group	19/30	60.3 ± 20.2	57.8 ± 20.5	*.686*
Been to rehabilitation stay	14/35	42.9 ± 21.1	65.1 ± 16.2	*.000*
Participated in disease-specific coping courses	18/31	57.2 ± 18.4	59.7 ± 21.4	*.686*
Received aids (i.e. wheelchair, taxi-drive to school)	17/32	57.4 ±14.6	59.5 ± 22.8	*.724*
Delayed school progression	37/12	56.9 ± 19.5	64.6 ± 22.2	*.256*
Participate in leisure activity	21/41	60.0 ± 17.2	53.7 ± 20.6	*.232*

Supplemental Table 4: HRQoL as measured by PedsQL4.0– social functioning dimension, variables from questionnaires and data collected from patient journals.

PedsQL4.0 dimension- Social functioning	**N**	**Mean** (SD)	**Mean** (SD)	
	Yes/No	**Yes**	**No**	***p***
Factors before diagnosis				
Using medications before diagnosis	17/30	60.9 ± 18.7	73.2 ± 13.2	*.012*
School attendance before diagnosis < 50 %	39/9	66.7 ± 16.4	77.8 ± 11.8	*.062*
Factors at diagnostic evaluation				
Diagnosed in outpatient clinic (not hospitalized)	38/9	69.2 ± 15.9	68.9 ± 17.6	*.958*
Joint consultation by medical and psychiatric units	22/20	68.9 ± 15.3	69.5 ± 13.2	*.886*
Child/adolescent psychiatry engaged in diagn. eval.	45/4	67.9 ± 16.1	76.3 ± 14.9	*.322*
Nurse engaged in diagnostic evaluation	14/35	66.4 ± 19.2	69.4 ± 14.8	*.559*
Nutritionist engaged in diagnostic evaluation	5/44	*72.0 ± 9.1	68.2 ± 16.6	*.618*
Physical therapist engaged in diagnostic evaluation	34/15	70.7 ± 12.8	63.7 ± 21.3	*.248*
Occupational therapist engaged in diagn. evaluation	31/18	70.3 ±13.2	65.6 ± 20.1	*.320*
Educator engaged in diagnostic evaluation	7/42	75.0 ± 18.3	67.5 ± 15.6	*.255*
Social worker engaged in diagnostic evaluation	2/47	72.5 ± 10.6	68.4 ± 16.3	.727
Collaborative meeting with primary health care held	9/9	63.3 ± 26.5	74.4 ± 9.2	*.262*

Factors after diagnosis				
Individualized school schedule	30/19	67.3 ± 14.2	73.2 ± 13.5	*.160*
Individualized examination plan	17/32	67.1 ± 14.6	70.9 ± 13.8	*.364*
Home education by teacher	10/39	65.0 ± 17.5	70.8 ± 13.1	*.251*
Organized schooldays (with or without individualized school schedule)	43/6	68.7 ± 14.4	75.8 ± 10.2	*.250*
Support from schoolteacher	42/7	71.1 ± 12.9	60.7 ± 18.6	*.071*
Support from advisor in school	38/6	70.0 ± 13.8	59.2 ± 15.6	*.085*
Been seeing pedagogic-psychologic services	27/21	68.1 ± 15.5	72.4 ± 11.6	*.301*
Been seeing social worker	7/41	73.6 ± 11.8	68.2 ± 13.7	*.333*
Been seeing nutritionist	8/39	66.9 ± 11.3	70.0 ± 15.0	*.580*
Been seeing a psychiatric nurse or nurse	2/45	72.5 ± 10.6	68.7 ± 13.8	*.701*
Been seeing a school nurse	12/37	70.0 ± 8.3	69.5 ± 15.6	.909
Been to psycho motoric physical therapist	4/32	73.8 ± 8.5	69.8 ± 14.9	*.615*
Been to physical therapist	27/22	67.6 ± 12.4	72.0 ± 15.8	*.275*
Been seeing an occupational therapist	25/23	68.4 ± 12.8	69.6 ± 14.5	*.768*
Been to child and adolescent psychiatry	28/21	67.1 ± 14.9	72.9 ± 12.5	*.161*
Been seeing a general practitioner	37/11	68.9 ± 14.2	72.7 ± 14.0	*.439*
Responsibility group	19/30	70.3 ± 17.1	69.2 ± 12.0	*.793*
Been to rehabilitation stay	14/35	59.3 ± 14.4	73.7 ± 11.8	*.001*
Participated in disease-specific coping courses	18/31	69.4 ± 16.1	69.7 ± 13.0	*.956*
Received aids (i.e. wheelchair, taxi-drive to school)	17/32	67.1 ± 12.4	70.9 ± 14.9	*.364*
Delayed school progression	37/12	66.1 ± 12.7	80.4 ± 12.9	*.001*
Participate in leisure activity	21/41	75.2 ± 11.5	62.4 ± 15.9	*.002*

Supplemental Table 5: HRQoL as measured by PedsQL4.0 – school functioning dimension, variables from questionnaires and data collected from patient journals.

PedsQL4.0 dimension – School functioning	**N**	**Mean** (SD)	**Mean** (SD)	
	Yes/No	**Yes**	**No**	*p*
Factors before diagnosis				
Using medications before diagnosis	15/28	36.0 ± 26.0	47.5 ± 20.7	*.121*
School attendance before diagnosis < 50 %	37/8	39.3 ± 21.0	61.9 ± 23.7	*.010*
Factors at diagnostic point				
Diagnosed in outpatient clinic (not hospitalized)	35/8	48.1 ± 26.6	44.0 ± 21.8	*.645*
Joint consultation by medical and psychiatric units	18/20	41,9 ± 22.6	44.0 ± 21.7	*.777*
Child/adolescent psychiatry engaged in diagn. eval.	41/4	43.8 ± 23.0	38.8 ± 25.3	*.681*

Nurse engaged in diagnostic evaluation	14/31	37.5 ± 16.4	46.0 ± 25.2	*.187*
Nutritionist engaged in diagnostic evaluation	3/42	63.3 ± 12.6	41.9 ± 23.0	*.120*
Physical therapist engaged in diagnostic evaluation	31/14	44.7 ± 21.4	40.4 ± 26.8	*.565*
Occupational therapist engaged in diagn. evaluation	29/16	43.3 ± 21.3	43.4 ± 26.4	*.982*
Educator engaged in diagnostic evaluation	7/38	56.4 ± 16.0	40.9 ± 23.4	*.101*
Social worker engaged in diagnostic evaluation	2/43	35.0 ± 21.2	43.7 ± 23.2	*.605*
Collaborative meeting with primary health care held	9/8	39.4 ± 27.2	43.8 ± 26.2	*.745*
Factors after diagnosis				
Individualized school schedule	27/19	40.7 ± 19.2	47.6 ± 26.1	*.307*
Individualized examination plan	17/29	37.6 ± 19.5	47.1 ± 23.4	.169
Home education by teacher	10/36	38.5 ± 26.0	45.0 ± 21.3	*.421*
Organized schooldays (with or without individualized school schedule)	41/5	41.8 ± 21.7	58.0 ± 23.9	*.127*
Support from schoolteacher	39/7	45.8 ± 22.5	31.4 ± 17.5	*.118*
Support from advisor in school	35/6	43.6 ± 23.5	35.8 ± 19.1	*.451*
Been seeing pedagogic-psychologic services	26/19	39.4 ± 21.6	50.0 ± 22.7	*.119*
Been seeing social worker	7/38	36.4 ± 19.1	44.0 ± 22.3	*.409*
Been seeing nutritionist	8/36	34.4 ± 13.5	45.6 ± 24.0	*.213*
Been seeing a psychiatric nurse or nurse	2/42	50.0 ± 21.2	42.6 ± 22.3	*.649*
Been seeing a school nurse	12/34	36.3 ± 12.6	46.2 ± 24.4	*.082*
Been to psycho motoric physical therapist	4/31	55.0 ± 20.4	43.1 ± 22.2	*.318*
Been to physical therapist	25/21	37.2 ± 18.7	51.2 ± 24.2	*.032*
Been seeing an occupational therapist	23/22	41.1 ± 23.4	44.6 ± 20.5	*.601*
Been to child and adolescent psychiatry	25/21	43.8 ± 19.7	43.3 ± 25.5	*.945*
Been seeing a general practitioner	35/10	42.6 ± 21.6	47.5 ± 26.4	*.548*
Responsibility group	19/27	42.4 ± 21.9	44.4 ± 22.9	*.759*
Been to rehabilitation stay	13/33	38.5 ± 21.6	45.6 ± 22.5	*.333*
Participated in disease-specific coping courses	17/29	39.7 ± 23.1	45.9 ± 21.8	*.372*
Received aids (i.e. wheelchair, taxi-drive to school)	16/30	37.8 ± 20.0	46.7 ± 23.1	*.203*
Delayed school progression	34/12	38.1 ± 20.5	59.2 ± 20.4	*.004*
Participate in leisure activity	21/36	50.5 ± 19.5	35.7 ± 20.8	*.011*

Supplemental Table 6: HRQoL as measured by PedsQL4.0– psychosocial functioning dimension, variables from questionnaires and data collected from patient journals.

PedsQL4.0 dimension – Psychosocial functioning	N Yes/No	Mean (SD) Yes	Mean (SD) No	*p*
Factors before diagnosis				
Using medications before diagnosis	17/30	50.2 ± 19.6	60.5 ± 14.5	*.045*
School attendance before diagnosis < 50 %	39/9	54,2 ± 15,9	67,7 ± 17,7	*.029*
Factors at diagnostic evaluation				
Diagnosed in outpatient clinic (not hospitalized)	38/9	57.3 ± 15.6	59.7 ± 21.3	*.696*
Joint consultation by medical and psychiatric units	22/20	58.2 ± 16.3	56.8 ± 14.0	*.756*
Child/adolescent psychiatry engaged in diagn. eval.	45/4	57.1 ± 17.2	55.4 ± 13.6	*.854*
Nurse engaged in diagnostic evaluation	14/35	54.3 ± 16.2	58.0 ± 17.1	*.493*
Nutritionist engaged in diagnostic evaluation	5/44	70,3 ± 2,7	55,4 ± 17,1	*059*
Physical therapist engaged in diagnostic evaluation	34/15	59,8 ± 14,1	50,5 ± 20,8	*.075*
Occupational therapist engaged in diagn. evaluation	31/18	59,1 ± 14,5	53,2 ± 20,0	*.247*
Educator engaged in diagnostic evaluation	7/42	65.2 ± 15.0	55.5 ± 16.8	*.159*
Social worker engaged in diagnostic evaluation	2/47	61.7 ± 14.1	56.7 ± 17.0	*.688*
Collaborative meeting with primary health care held	9/9	50.7 ± 24.5	63.4 ± 13.2	*.191*
Factors after diagnosis				
Individualized school schedule	30/19	56.0 ± 15.2	60.3 ± 17.5	*.374*
Individualized examination plan	17/32	54.1 ± 13.8	59.6 ± 17.0	*.264*
Home education by teacher	10/39	54.2 ± 19.2	58.6 ± 15.3	*.445*
Organized schooldays (with or without individualized school schedule)	43/6	56.7 ± 16.4	64.9 ± 12.4	*.246*
Support from schoolteacher	42/7	59,8 ± 15,1	44,8 ± 16,8	*.020*
Support from advisor in school	38/6	58.4 ± 16.4	46.7 ± 15.2	*.106*
Been seeing pedagogic-psychologic services	27/21	56.0 ± 15.5	61.0 ± 16.3	*.285*
Been seeing social worker	7/41	55.5 ± 11.2	57.2 ± 16.2	*.786*
Been seeing nutritionist	8/39	53.3 ± 12.1	58.5 ± 17.2	*.425*
Been seeing a psychiatric nurse or nurse	2/45	59.2 ± 20.0	56.7 ± 15.6	*.831*
Been seeing a school nurse	12/37	55.8 ± 10.6	58.3 ± 17.6	*.653*
Been to psycho motoric physical therapist	4/32	58.3 ± 11.8	57.5 ± 17.1	*.928*
Been to physical therapist	27/22	55,5 ± 14,1	60,3 ± 18,1	*.298*
Been seeing an occupational therapist	25/23	57.3 ± 14.9	56.6 ± 16.3	*.864*
Been to child and adolescent psychiatry	28/21	55.9 ± 16.4	60.1 ± 15.6	*.369*
Been seeing a general practitioner	37/11	56.4 ± 15.6	62.2 ± 18.4	*.303*
Responsibility group	19/30	57.6 ± 17.4	57.7 ± 15.4	*.990*

Been to rehabilitation stay	14/35	47.1 ± 16.8	61,9 ± 13,9	*.003*
Participated in disease-specific coping courses	18/31	56.0 ± 16.7	58.7 ± 15.9	*.578*
Received aids (i.e. wheelchair, taxi-drive to school)	17/32	54.7 ± 12.2	59.3 ± 17.7	*.344*
Delayed school progression	37/12	54.3 ± 14.3	68.1 ± 17.3	*.008*
Participate in leisure activity	21/41	61,9 ± 13,2	51,7 ± 16,6	*.017*

Paper II

BMJ Open

Factors related to educational adaptations and social life at school experienced by young people with CFS/ME: a qualitative study

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ABSTRACT

Objectives To explore factors perceived as positive or negative among young people with chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) in relation to school and everyday life.

Design A qualitative study with semistructured individual interviews performed at the local hospital or at the informants' homes between September 2017 and January 2018, with an additional telephone interview to collect data on experiences from the COVID-19 pandemic, conducted in September 2020. Data were analysed using a grounded theory approach.

Setting The informants were recruited from two university hospitals that offer interdisciplinary assessments of young people with CFS/ME from various parts of Norway.

Participants Five males and 13 females aged 13–21 years with CFS/ME diagnosed 3–56 months prior to the interviews participated.

Results The informants were concerned about a lack of educational adaptations and missed social life at school. Educational and social adaptations could improve schooling and health among young people with CFS/ME. Negative experiences were related to a lack of knowledge about CFS/ME among school personnel and young people's difficulties to limit activities. Online teaching as experienced during the COVID-19 pandemic was described as positive both for education and social life.

Conclusions Young people with CFS/ME can benefit from better educational adaptations and increased social interaction with peers. From the participants' view, factors that limit learning and socialisation include a lack of knowledge about CFS/ME among teachers and school personnel, expectations from teachers of doing more than they could manage at school, feeling alone coping with the disease and not recognising their own limitations regarding what they are able to do. Suggested factors perceived to enhance learning and socialisation were a better understanding of the disease among school personnel and peers, suitable educational adaptations and being able to socialise with peers.

Strengths and limitations of this study

- The assessment of young people with chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) was similar in the two university hospitals from where informants were recruited.
- All interviews were performed by a person who had cared for young people with CFS/ME to ensure attention to the specific needs for this patient group.
- A limitation to the study was that precise information about what adaptations the young people had received at school was not provided.
- A limitation was also that findings may not be directly transferable to countries with other educational and healthcare systems as the study was set in Norway.
- A third limitation was that the parents' involvement in adaptations at school was not explored.

BACKGROUND

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) is a disease characterised by general fatigue and post-exertional malaise (PEM) for the duration of at least 3 months.[1] CFS/ME occurs three to four times more frequently in females than males with an estimated prevalence of 0.1%–1.0%.[2] The incidence rate of CFS/ME among Norwegian students is 43 out of 100 000.[2] The aetiology of the condition is unknown and there is currently no curative treatment.[3] Young people with CFS/ME usually have impaired physical and cognitive functioning, which may lead to disruption of education, as well as social and physical activities during a critical period of life.[4 5] Depressive symptoms are common, possibly due to the loss of education and social interaction,[6] and a lack of awareness about the disease may cause anxiety.[7] Young people diagnosed with CFS/ME often find it difficult to limit activities.[4] They tend to put too much energy into activities like schooling, potentially leading to increased fatigue.[8] Previous studies found that young people with CFS/ME can benefit from educational adaptations and advice on

how to master school and everyday life.[5 9 10] Knowledge about which educational and social adaptations young people with CFS/ME find useful is still limited.

In a previous study, we found that good relationships with school and teachers were associated with higher levels of health-related quality of life (HRQoL) in young people with CFS/ME.[11] The aim of the present study was to explore factors perceived as positive or negative among young people with CFS/ME in relation to school and everyday life.

METHODS

Design

A qualitative study using semistructured individual interviews was conducted between September 2017 and January 2018. Supplementary interviews related to the new situation in schools created by the COVID-19 pandemic were conducted in September 2020.

Setting

The informants were recruited from two university hospitals that offer interdisciplinary assessments of young people with CFS/ME from various parts of Norway. After the assessment, medical doctors, occupational therapists, and specialised nurses usually communicate with primary healthcare providers and school personnel to inform them about the disease and educational or other adaptations the individuals might need.

In Norway, most schools are public with compulsory schooling for 10 years and with a right to complete additional 3 years of high school before the age of 24 years. At high school, students can choose between academic and vocational tracks. It is mandatory for all schools regardless of level to assess individual needs and adapt education. To obtain a high school diploma, most students must attend school 85% or more of the time. With a doctor's declaration documenting illness as the primary cause of school absence, exemption to this limit is possible. During the COVID-19 pandemic, schools offered teaching via online platforms beginning from March 2020.

Informants and recruitment

Informants were recruited by mail for the current study and a preceding HRQoL study.[11] Inclusion criteria were (1) a diagnosis of CFS/ME from St Olav's Hospital, Trondheim University Hospital or Oslo University Hospital in Norway and (2) age 12–18 years at the time of diagnosis. Exclusion criteria were (1) inability to understand Norwegian or (2) not able to participate in interviews. Among 33 informants contacted, 18 consented to participate in the interviews. Reasons for not participating were not wanting to or being able to participate in interviews.

The first author contacted all consenting participants consecutively after replying to the HRQoL study. All informants were diagnosed with CFS/ME according to Jason's paediatric diagnostic criteria.[12] Informants were included until preliminary analysed data were repeated by new informants and saturation was considered achieved.[13] Three of the informants had recently entered university. However, they primarily talked about their experiences from secondary and high school. Three of the informants had previously met the interviewer in a meeting or course at one of the hospitals.

Data collection and interview guide

The first author, a female PhD student and specialised nurse, interviewed all informants face-to-face. All the informants were offered interviews at home to avoid potential worsening of fatigue due to travel. Fourteen of the interviews were completed at the local hospital in a regular office, with comfortable chairs and dim light. Four informants were interviewed at home. Due to age and the potential negative cognitive impact of the disease, informants could bring a parent to the interview, which 11 of the informants did. The informants were told that they could pause or stop the interview at any time. Parents were informed that they should only comment on the informant's or interviewer's request.

The semistructured interview guide was developed by a group consisting of two medical doctors, two psychiatrists, one health economist and a specialised nurse. All were experienced with the patient group. The questions were based on previous knowledge about perceived challenges for young people with CFS/ME, especially factors important for education. Subsequently, the questions were discussed by the research group and informed by previous research. The interview guide consisted of two parts: the first part addressed being fatigued and the second part addressed healthcare and support from school, family and friends. The central questions were about 'How has your school curriculum been adjusted and how have the adjustments worked out for you?' and 'How do people you relate to perceive your disease?'. The interviewer did not have any information about provided adaptations for the informants from school personnel or healthcare providers. Interviews were audio recorded and lasted between 59 and 116 min. A pilot telephone interview with a young female with CFS/ME was performed. Comments were considered and resulted in minor changes to the questions, order of questions and a few additional questions.

The COVID-19 pandemic led to a new way of schooling for all young people in Norway. To study experiences from the COVID-19 pandemic, the first author conducted supplementary interviews by phone in September 2020. The central questions were about how the informants perceived the switch to online schooling and altered contact with school following the COVID-19 pandemic. Sixteen of the 18 informants participated in the supplementary interview.

Data analysis

The interviews were consecutively transcribed verbatim by the first author, and analyses were based on a grounded theory approach.[14–16]

Data were initially open coded by the first author into seven categories: (1) health, (2) own perception and acceptance of the disease, (3) others' perception and acceptance of the disease, (4) healthcare and support, (5) school adaptations, (6) emotional challenges, and (7) loss and sorrow. The open coding showed differences in how the participants experienced school adaptations, and educational adaptation was the main concern emerging from the data. Subsequently, the main concern was axially coded with the seven categories from open coding. In axial coding, own and others' perception and acceptance of the disease (categories 2 and 3) were considered important to how the need of school adaptations was resolved. These two categories were leading when data were selectively coded in new categories and described in a storyline and a conditional matrix. Following selective coding, loneliness emerged as a second important concern among the informants, where school contact seemed important to resolve the main concerns. Selectively coded data were passed from the first author to the coauthors. A research group at the university read and gave their view and perspectives to the preliminary results of the analysis.

Patient and public involvement

The patient perspective was included in the planning of the project by obtaining approval from the patient organisation. A patient representative was involved in the conduct of the study by commenting on the interview guide after participating in pilot testing.

RESULTS

A total of 18 informants, 5 males and 13 females, aged 13–21 (median age 19) years were interviewed (table 1). The informants were diagnosed with CFS/ME 3–56 months prior to the interviews, and two of them had recovered. Ten of the 18 informants attended more than half of the classes in the 4-week period before the interview. Five out of 18 informants did not attend school at all in the 4-week period before the interview, but three had contact with school personnel and received home assignments.

Table 1 Characteristics of informants

Characteristic	N
Gender	
Female	13
Male	5
School level	
Secondary school	5
High school	10
University	3
School attendance	
0%	5
1%–50%	3
51%–100%	10

School level and school attendance are at the time of the interview.

The participants experienced a range of symptoms. As expected, the most common symptom was fatigue, which typically worsened if they pushed themselves to do more than they could manage. Furthermore, insomnia/unrefreshing sleep, headache, joint and muscle pain, and hypersensitivity to light, sounds, or smells were recorded. Other symptoms mentioned were neurocognitive manifestations (ie, memory and concentration difficulties), autonomic, neuroendocrine and immune manifestations. Symptoms could vary and fluctuate over time. They had all been advised by healthcare providers to follow a daily plan with fewer activities. Most of them experienced this as useful, but two said that a daily plan did not work out for them. Some received assistive devices like noise-cancelling headphones, special sunglasses or a wheelchair.

Lack of adaptations at school was a main concern among the informants. Management of the school day seemed to depend on how the young people themselves and others perceived and accepted their disease. Many felt that they were not understood by school personnel, and some said it was difficult to know how to initiate and handle school adaptations. In addition, they felt lonely because they were not able to participate as normal in social life with peers.

Words and phrases like 'terrible', 'tough' and 'locked in and trapped' were used by informants to describe how it was to live with CFS/ME. They said they felt angry, irritated and sad about not being able to go back to school and their regular social life. Some of their everyday life experiences from living with CFS/ME are shown in figure 1.

Fewer spoke about negative experiences regarding school adaptations among the most recently diagnosed than those who had been diagnosed years ago. Factors that the informants perceived as positive or negative in relation to school were categorised into the following four themes: *Educational adaptations and challenges, Focus on what you can do or focus on the illness, Social life and support,* and *Adaptations following the COVID-19 pandemic.*

Educational adaptations and challenges

Adaptations that worked

Some informants said that they were taken seriously by their teachers and received adaptations to accommodate their needs. One example was an educational plan with fewer lessons where the balance between activity and rest during the school day was taken into consideration. Some received fewer tests, or tests at home, and an adjusted overall academic progression plan. They had an opportunity to socialise with peers at school, and they were supported by teachers, counsellors, and school nurses. Two participants who attended special education schools received all these adaptations and were content with their

I was used to keeping busy. (…) I was living as normal. (…). It was very hard because I could not go to school. (…) I will soon be back (…) two more months and you`re well (…) But after we stopped thinking [that you`re well after two months], it automatically became better." (YP11, HS).

In the informant's opinion, there was nothing they could do to improve the fatigue, other than rest and try not to wear out. Getting a diagnosis took too long, and some were not taken seriously by health care providers in the beginning. They commonly experienced fluctuating symptoms; they tended to feel better during holidays when they had no schoolwork, and usually became worse after going back to school. Close family members and close friends were always supportive, while more distant family members and friends had difficulties understanding their situation. It was difficult to explain to others how they felt as there were no visible signs of disease. No one could fully understand how it was living with CFS/ME without having experienced it.

«Just now I am not much at school, and I think that is a huge problem. (…). It`s hard to make plans (…), I always have to say no" (YP1, HS).

Figure 1 Everyday life experiences from living with CFS/ME. CFS/ME, chronic fatigue syndrome/myalgic encephalomyelitis.

school situation. They especially valued the good atmosphere, alternative educational tasks, and close contact with teachers and counsellors. Informants who received adaptations that worked talked about more regular school attendance than other informants.

> I have the same plan as the rest of the class, but I have the possibility to extend deadlines or miss out on some projects (…) because it is too demanding, but I am able to attend my class, and that is also what I want. (YP12, high school (HS))

A struggle to get educational adaptations

Regular school days were difficult to handle for all the informants in the beginning of their disease. Some said that they were not listened to and had to struggle to get the educational adaptations they needed. Some even studied at home without any communication with a teacher and had to ask friends to provide notes from lessons.

A plan for school attendance was useful, yet many experienced that the plan often included more items than they could cope with. Overambitious plans could be made by teachers, but the informants also said that it could be difficult for themselves to recognise their own limitations. They would overestimate their capabilities to attend school, resulting in worsening of fatigue, PEM and increased school absence.

> Yes, I believe it's just a matter of knowing your limitations, like when I have a very good day, I can't use all the energy, because then I know that I will have a bad day tomorrow somehow (…). (YP7, HS)

Some of the informants said they felt overlooked and disbelieved by their teacher. If they did not manage to follow the plan or if they missed school, teachers questioned the reason for their absence and did not contact them or update plans as previously promised. Often this meant that their parents were left with the responsibility to request meetings with the school to update plans.

> Presently I don't attend school (…). Some [teachers] understood, while some didn't understand completely, and some didn't understand at all. (…) Those who understood some, promised a lot, and then it wasn't possible to carry through. (YP5, HS)

The informants said they wanted to go to school but were not always able. They said it felt like a total defeat when they realised that they could not progress through school normally. Two of the informants who eventually had started in special education schools said it was hard to accept for them at first, after a while they felt it was the best thing that happened to them. Now they could attend school even on a bad day and get the help they needed.

> So, returning [to school], just for a bit at least, that is really important. It's more about considering that the school takes an initiative. (…) I haven't heard from them since long before Christmas. (YP10, secondary school (SS))

Before being diagnosed, several informants said they were not always believed by their teachers thus, they received no adaptations for education nor social life at school. They did not manage to keep up with the rest of the class, and some of them started feeling stupid. One said that this led to low self-esteem and resistance towards attending school. After receiving a diagnosis, many said it was easier to be believed by teachers. Others said that it did not lead to improvements, even after healthcare providers had informed the teachers. Some of the informants said that they perceived the lack of understanding from teachers to be caused by a lack of knowledge about CFS/ME.

> Before we got a really good collaboration with school there were uncertainties in a way, and particularly before I was diagnosed, (…) it was more like I come [to school] when I come, and the teachers choose not to ask. (YP12, SS)

Focus on what you can do or focus on the illness

Some informants said that if they focused on their fatigue and what they no longer could do, it made them feel depressed. Focusing on what they were able to do, accepting life as it was, staying positive minded and socialising when they were able made them feel better. Diversions like television or a pet were also helpful for some. One said that for a while, it had worked well not to think about the help she hoped for but did not get from the school.

> If I must consider how difficult and hard and strenuous and tiring and hurtful and miserable everything really is, with my disease and my diagnosis and how I experience this. If I focus on that, so, you know, I simply don't think I would stay alive. (YP11, HS)

The informants said they mostly focused on the present. They did not like to think about the future because of the uncertainties concerning the duration of the disease and the difficulties they had experienced with educational adaptations. One feared that the future could bring more experiences of being alone with her disease challenges. To think about major transitions between school were especially worrying, as they previously had experienced those changes could lead to worsening of the disease. If good communication between educators was assured, major transitions felt safe.

> Standing there alone, is what I fear, because I have been [alone], and I remember how terrible it was. I don't want to stand alone, because I have worked hard for the things I have now, (…) the transition, I fear that, it can go terribly wrong (…) I have no idea. I don't know my opportunities. I don't know who can be there, what titles, or functions or I don't know, (….) and that has been a huge problem through my illness, that I don't know. I don't know where to get information. I don't know who I can ask. (YP11, HS)

Social life and support

All informants said that they missed meeting peers. Socialising with peers was difficult both due to fatigue and to school absence. Some were able to prioritise socialising during the school day, that is, with a plan that included attending one or more breaks with peers. Others said that school did not prioritise social interactions with peers in the educational plans. Some had friends they met with regularly in their leisure time, while for others it could go months without socialising with peers. At home they socialised with family when they managed. Some feared they would enter adult life without having developed socially among peers either at school nor in everyday life.

> (…) In many ways I haven't been able to develop socially like other teenagers, because I don't have the freedom to go where I want or do what I want (…). (YP1, HS)

The informants noticed that it was difficult for family and friends to understand the implications of the disease. In the beginning, family members could push them to school without knowing when they had to stop. This eventually improved, and the participants experienced that close family members accepted and respected them and were very supportive. The informants had experienced that close friends tried their best to understand and to support them, and that those friends could feel powerless because there was no improvement. Some friends who had trouble believing or accepting withdrew and eventually disappeared.

Someone to talk to

Many believed that family, friends, teachers and school nurses could all help them to cope with their disease. Most informants talked with close family and friends about frustrations regarding the disease. Only a few informants said they talked to their teachers or school nurses, and the reason for those who did not was that they expected the school nurses or teachers would not understand. Subsequently, this made them feel alone regarding how to handle the disease and challenges at school. Some suggested the potential helpfulness in a school counsellor who understood their situation, who could help them if the disease worsened and counsel them regarding future education and realistic job opportunities.

> Maybe someone can help me plan in a way, maybe about the future, when I leave high school. (…) a type of counselor, maybe a school counselor or something or someone and just see opportunities and what I can do, so one doesn't feel that this is what I am going to do the rest of my life. (YP15, university)

Online teaching might be a useful adaptation for young people with CFS/ME

Online teaching became a necessity during the COVID-19 pandemic. The informants described online teaching as helpful because everyone had to stay at home, plans had to be made, and thus, the informants could participate on equal footing with their peers. One informant said that online tasks, often with a completion deadline set later than before, gave him the freedom to do schoolwork whenever he managed to, so he felt more in control. A second informant said that online teaching had enabled her to complete the school year even though she was housebound. The informants reported that teachers who did not manage to adapt teaching to their needs previously were able to do so during the COVID-19 pandemic.

> Online teaching was almost the same as meeting one-on-one at home. It was in a way nice that everybody was in the same situation with online teaching. Everyone was stuck together. Doesn't feel so different, I was like most others. The opportunity of online teaching would be better if I had it from first year of high school. (…). I think it's a good solution for the chronically ill. (YP1, HS)

Communicating online with teachers through chat was described as easier and more direct than the previous use of email. One informant thought that if she had been taught via online platforms earlier, she could have kept in contact with peers and teachers more directly, and that this would probably have made it easier to attend physical school again later. However, some said they still preferred to meet physically with teachers and peers rather than via online platforms. These participants felt it was easier to

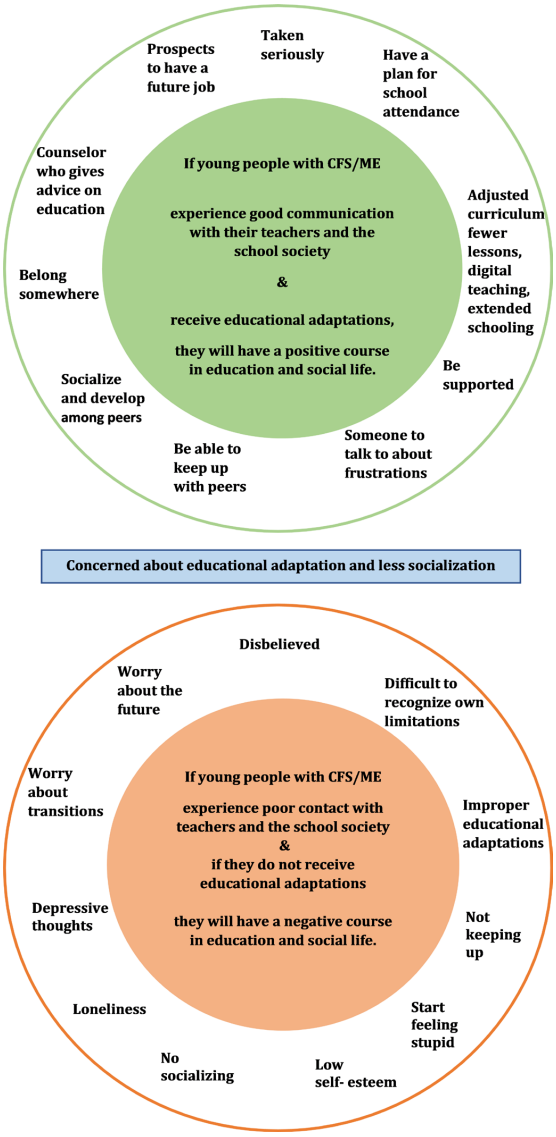

Figure 2 How the informants experienced that their main concerns, for example, adaptation of education and social life was resolved and how the resolving factors resulted in positive (green) or negative (red) course. CFS/ME, chronic fatigue syndrome/myalgic encephalomyelitis.

follow lessons when they met physically at school and had the possibility to ask the teachers questions more directly.

> I liked better to meet people. I was fine being at home. It was a bit more difficult because the teacher can't come over and explain to you. (YP4, SS)

In figure 2, the overall results are summarised in a twofold conditional matrix. A conditional matrix can be visualised as a set of concentric circles with actions and interactions at the centre, and where each level of circles corresponds to a different unit of influence on these actions. In this conditional matrix, the inner circles show how the participants experienced interaction with their teachers, how teachers acted to adapt education and social life at school for them, and how this resulted in a positive or negative educational course. The outer circles show the factors that were experienced as positive or negative regarding education and social life at school, and how the factors impacted the educational course.

DISCUSSION

This study explored factors perceived as positive or negative among young people with CFS/ME in relation to school and everyday life. The main findings were the informants' concerns about educational adaptations and social life at school. Positive factors were appropriate measures that could lead to improved learning and a better social life. On the other hand, a lack of measures or maladaptive measures could limit learning and in the worst case cause dropout from school. A novel and potentially important finding was that online teaching improved learning and made young people with CFS/ME feel more socially connected with both teachers and peers.

A strength of this study was that assessment and follow-up of young people with CFS/ME were similar in the two university hospitals included. All interviews were performed by one person experienced with young people with CFS/ME to ensure attention to the specific needs for this patient group. It was a limitation to the study that we did not register precisely what adaptations the young people had received at school. Furthermore, parents' involvement in adaptations was not explored. Another limitation was that the findings may not be directly transferable to countries with other educational and healthcare systems than Norway. This was the first study in Norway to explore perceived factors related to school functioning among young people with CFS/ME.

Some of the informants had experienced satisfying educational and social adaptations at school and managed to maintain their education. Factors related to satisfaction were typically being taken seriously, good communication with school, and individually tailored educational and social adaptations. Most young people with CFS/ME manage to keep up with their peers if they are given an adjusted curriculum.[3] Educational adaptations responding to the needs of young people with CFS/ME require that health and school personnel have knowledge about how CFS/ME impact learning,[3] and how to address the need for adaptation in school.[4] This requires sound scientific and practical knowledge among healthcare providers, and that information on individual educational and social challenges is communicated to school personnel.[17 18] This includes the important engagement from occupational therapists in identification of potential obstacles, solutions and subsequent advice to teachers

on individualised adaptations.[19] Teachers' genuine concern about their students' needs is valuable, nevertheless, attitudes towards CFS/ME might impact experiences in students with CFS/ME.[17] Advice from healthcare providers is usually necessary to prevent a course of intuitive trying and failing when adapting education for young people with CFS/ME.[20] Teachers also need acceptance and resources from their leaders to be able to adapt education adequately to the student's needs.[21] Conversely, if the teachers do not see any reason for adapting the education level, the school leadership should take actions to secure individual educational adaptations for students with CFS/ME.[21] The current study confirms that adequate and individualised educational and social adaptations at school may facilitate school progression for young people with CFS/ME.

Other informants described several negative factors related to educational and social adaptations in school. Negative factors included disbelief and distrust from teachers, overambitious educational plans and difficulties in recognising their own limitations. Not all schools have previous experiences of students with CFS/ME. In addition, the adolescents might try to conceal their symptoms when they attend school, potentially making it difficult for teachers to understand their adaptation needs.[1 3] Also, the uncertainties surrounding CFS/ME diagnosis and especially CFS/ME aetiology could further contribute to a lack of understanding and disbelief from schools.[17] This may lead to overambitious plans for school attendance.[4] One reason for why overambitious or inadequate plans are made can be to reduce the negative consequences for healthy students.[21]

Nevertheless, in Norway, schools are required by law to adapt education for young people with chronic health conditions. There is however a gap between ambitions and realities when adaptations of education for young people with chronic health conditions are managed in mainstream classes.[22] The current finding confirmed that the informants' experienced that promised adaptations were not always possible to carry through. Special classes have been described as preferable for young people with other chronic health conditions.[23] This was also preferred by some informants in the current study.

How the interdisciplinary management of adaptations for young people with chronic health conditions in mainstream classes is handled and how they work out is poorly explored. Thus, young people with CFS/ME might not be the only ones who struggle with educational and social adaptations at school. It is previously explored how healthcare providers' support schools regarding the needs in young people with chronic health conditions,[24] and it is found that social teachers can facilitate social connectedness with peers for young people with disabilities.[25]

Some informants found it difficult to recognise their own limitations regarding activities at school and suggested that an advisor perhaps could help them to find a more appropriate activity level. Young people with CFS/ME commonly spend most of their energy trying to keep up with schoolwork early in their disease,[8] motivated by their previous experience of being able to participate and master school when healthy.[26] The need of young people with CFS/ME for educational adaptations is previously described as important.[4 27–29] However, educational adaptations are often delayed due to the long time it takes to establish a CFS/ME diagnosis.[29] The current study confirms that young people with CFS/ME often lack early adaptations of education. It also confirms that young people with CFS/ME appreciate guidance on how to adjust to their limitations to ensure the best possible school progression and socialization with peers.

Disbelief from teachers and missed opportunities to develop both academically and socially among peers make young people with CFS/ME feel lonely.[4] An uncertain prognosis, increased emotional vulnerability, and lack of awareness and acceptance of CFS/ME also impact their mood.[6 7] It is challenging for young people with CFS/ME to accept that loneliness and dependency on close relatives often become the new everyday life instead of developing independence and their own social network.[26 30] The change in everyday life due to CFS/ME has previously been found to cause fragility and vulnerability as well as undesired emotions like irritability, worry, anxiety, sadness and depression.[4 26 31] Supportive relations and socialising in school are found to enhance understanding of the identity and loneliness challenges they meet.[4 7] The classroom might be the only place young people with CFS/ME meet with peers, and thus, social adaptation in school to allow for this is highly valued.[3] The current study confirms that young people with CFS/ME find it challenging to accept their new situation, that they experience loneliness and that this impacts how they feel.

Online teaching was perceived as helpful by most informants. It put them on equal footing with peers, and they experienced increased contact with teachers and peers. There are, to our knowledge, no studies examining experiences with online teaching for young people with CFS/ME, but it has previously been found that they can benefit from online healthcare treatment programmes.[32] Students with concentration impairments have benefited from being able to study without distractions from the physical classroom environment and from being able to study at their own pace.[33] Furthermore, previous studies have found that young people with CFS/ME find ways to adapt and maintain a sense of normality through online connectedness.[34] One study found that young people with CFS/ME do not perform academically to their full potential despite receiving external educational adaptation and support. Poor school-related quality of life and poor connectedness with school place them at risk of developing long-term maladjustment of cognitive, academic and social skills.[35] The current study adds to this by showing that young people with CFS/ME may benefit from online teaching both academically and socially.

Clear information on adaptive measures from healthcare to schools is an important factor to increase

knowledge about CFS/ME in schools. Young people with CFS/ME need early guidance and adaptation of education to adjust to their limitations. Online teaching may be an important factor both to improve learning and to improve social connectedness for young people with CFS/ME. Further research should focus on preventing loss of function among young people during the period before a CFS/ME diagnosis, in specific in regard to providing educational and social adaptations for young people with CFS/ME.

CONCLUSION

Young people with CFS/ME can benefit from better educational adaptations and increased social interaction with peers. According to young people with CFS/ME, factors that limit learning and socialisation include a lack of knowledge about CFS/ME among teachers, school personnel and in the educational system. Young people with CFS/ME feel alone coping with the disease and how to recognise their own limitations regarding what they are able to do. Factors that may facilitate learning and socialisation are a better understanding of the disease among teachers, school personnel and peers; suitable educational adaptations and being able to socialise with peers.

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Contributors WAS collected, analysed and interpreted the data, and was the main author of the manuscript. TBR supervised the project, analysed and interpreted the data, contributed to writing of the manuscript, and was the guarantor of this work. THN supervised interpretation of data and contributed to writing of the manuscript. IBH facilitated and supervised the data collection from Oslo University Hospital. All authors read and approved the final manuscript.

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Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

Patient consent for publication Obtained.

Ethics approval All procedures performed were in accordance with ethical standards and approved by the Regional Ethical Committee for medical and health profession research in South-East Norway (REK 2017/749) and performed according to the Declaration of Helsinki. Specifically considered protection for the participants was made. Participants were allowed to have their interviews at home to avoid worsening from travelling, to stop the interview if they needed, to voluntarily bring a parent, and to be seated in comfortable chairs and with dim light during the interview. The informants were also offered supportive healthcare after the interviews. Informed consent was obtained from all participants included in the study. Parents signed on behalf of informants below legal age (16 years). All informants received verbal and written information about the study. Informed consent obtained from all individual participants included consent for publication of anonymised data.

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Data availability statement Data are available upon reasonable request. The datasets generated and analysed during the current study are not publicly available. The raw data supporting the findings of the manuscript can be found at the Children's Clinic, St Olav's Hospital, Trondheim University Hospital, Trondheim, Norway. Due to regulations of the Regional Ethical Committee for medical and health profession research in Norway, REK, the anonymity of the informants must be secured. In the raw data, it is possible to identify the informants, and restrictions therefore apply to the availability of these data. Reasonable request concerning the data can be sent to the corresponding author.

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Paper III

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ORIGINAL RESEARCH
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Experiences Among School Personnel and School Nurses on Educational Adaptations for Students With CFS/ME: A Qualitative Interview Study

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Introduction: Chronic fatigue syndrome (CFS/ME) is a disabling disease severely impacting school attendance, education, and social life in young students. Uncertainties surrounding CFS/ME etiology may impact the interpretation of CFS/ME in schools. Thus, school personnel need information from health care providers to make adequate adaptations to education and social life at school for these students.

Objectives: To explore teachers, counselors, and school nurses' experiences with adapting education for students with CFS/ME aged 13–19 in secondary and high schools.

Design: A qualitative study with focus group interviews and individual interviews performed face-to-face or digitally between November 2020 and March 2021. Data were analyzed using Systematic text condensation.

Participants: Six teachers, two counselors, and four school nurses in secondary and high school participated.

Results: Adapting education for students with CFS/ME was challenging, especially before the students received a diagnosis. The challenges were related to identifying the students' adaptational needs, maintaining a teacher-student relationship due to school absence, difficulties in maintaining continuity of education, and uncertainty regarding the diagnosis. Successful adaptations were related to quickly reacting to school absence, early referral to educational, psychological services, a close collaboration with the school management, and the development of digital teaching for students with CFS/ME. Interdisciplinary collaboration and a clear, constructive plan with adaptive measures, including maintained teacher-student communication and educational and social adaptations, may be useful in preventing the losses, young people, with CFS/ME experience.

Conclusion: Early interdisciplinary collaboration to adapt education and social life at school for students with CFS/ME, may support teachers, counselors, and school nurses in their efforts to adapt education and prevent losses related to academic and social development in students with CFS/ME.

Keywords: chronic fatigue syndrome, education, social, school teachers, counselors, school nurse

INTRODUCTION

Young people's chronic health conditions may impact their health-related quality of life, academic performance, and social development (1–3). Identification of disease-related impairments with subsequent needs of educational adaptations is essential to ensure that these young people attend and continue schooling (4). Usually, the teacher is responsible for making the necessary adaptations to achieve this (5). Common adaptations include alternative tasks and home tasks as required, maintaining contact between teacher and student during school absence, and ensuring appropriate information exchange between teachers, parents, and health care providers. Management of these adaptations can be time-consuming for teachers, especially if they have several students with different chronic health conditions in their classes (6).

Chronic fatigue syndrome (CFS/ME) is a disabling disease with fatigue as the main symptom and a frequent worsening of symptoms after physical and cognitive activities, referred to as post-exertional malaise (PEM) (7, 8). Other symptoms are unrefreshing sleep, pain, cognitive impairments, and autonomic, neuroendocrine, or immune manifestations (7). The severity of CFS/ME symptoms differs in mild, moderate, severe, or very severe degrees, and may fluctuate along with the symptoms (8). The prevalence of CFS/ME is between 0.2 and 1.0% in young people (9, 10). Recently, it was found that post-COVID-19 might trigger CFS/ME in adolescents and young adults (11).

CFS/ME cause long-term school absence among young people (8, 12, 13), and for instance, school attendance fluctuates along with the severity of symptoms (14). Young people with CFS/ME suffer from physical, cognitive, and social impairments with severe impact on education and social life, and they need individual and flexible adaptations to participate in education and social life at school (1, 15–17). Adequate adaptations for students with CFS/ME are, i.e., later attendance, an opportunity to rest during the school day, fewer lessons, prolonged school progression, and facilitations to socialize with peers (1, 15). However, some students with CFS/ME are bedbound, socially isolated, and receive little help from their school (18).

Uncertainty surrounding CFS/ME etiology differs CFS/ME from other chronic health conditions (19). This uncertainty may impact the interpretation of the disease at school, and for this reason, the adaptations students with CFS/ME are offered may vary (20). Teachers need information and guidance from health care providers to successfully integrate students with CFS/ME in mainstream classes (4, 21). The school nurse may be an essential link between students, parents, teachers, general practitioners, and specialist health services regarding health needs for the students (22). Still, there is little knowledge about how school personnel and school nurses experience adapting education for students with CFS/ME.

Therefore, the aim of the study was to explore teachers, counselors, and school nurses' experiences with adaptations of education for students with CFS/ME aged 13–19 in secondary and high school.

METHODS

This was a qualitative study with a focus group and individual interviews conducted between November 2020 and March 2021. The study applied a phenomenological approach by focusing on the participants' lived experiences (23).

Reflexivity

The authors were one nurse and one medical doctor experienced in assessing young people with CFS/ME in a hospital and a third researcher with experience as a nurse but no experience with young people with CFS/ME. All authors had experience with qualitative methods. Two pedagogues in secondary and high school gave input to the interview guide to limit the influence of the authors' preconceptions on the interview guide.

Setting

In Norway, young people with CFS/ME under age 18 receive a diagnosis from specialist health care. Before CFS/ME diagnosis, the assessment includes thorough physiological, psychological, and social assessment and exclusion of other diagnoses. Generally, young people with CFS/ME are students in mainstream classes both before and after diagnosis. After diagnosis, school personnel and school nurses receive information about educational and social adaptations from health care providers. In the COVID-19 pandemic, the schools in Norway were locked down for physical attendance and offered digital education from March to June 2020.

Informants and Recruitment

The sampling strategy aimed to include participants from geographically spread schools, including both rural and urban districts.

The study aimed to include at least 24–32 participants divided into four or five focus group interviews. As the COVID-19 pandemic made restrictions to meet in groups, and inclusion thus became challenging, it was decided to switch to individual interviews after two focus group interviews.

Invitations to participate in the study were sent by e-mail to 18 secondary schools, 15 high schools, and 12 school nurses in

TABLE 1 | Characteristics of the informants.

Characteristics	*N* Tot (F/M)
Occupation	
Teacher	5 (3/2)
Teacher with leadership responsibility	1 (1/0)
Counselor in school	1 (1/0)
Counselor in EPS[a]	2[b] (2/0)
School nurse	4 (4/0)
Employment	
Employment in secondary school	9 (7/2)
Employment in high school	3 (3/0)
Participants experienced with 2 or more students with CFS/ME	9 (8/1)

F, Female; M, Male.
[a]EPS, educational psychological counseling services.
[b]One teacher had previous experience from EPS.

Mid-Norway. Inclusion criteria were being a teacher, counselor, or school nurse in secondary or high school and having previous or current contact with students with CFS/ME.

The first author contacted those who responded positively to participate with additional information about the study and to set a time for the interview. The study included four school nurses, six teachers, and two counselors from seven different schools in both rural and urban districts (**Table 1**).

Data Collection and Interview Guide

The first author, a female Ph.D.-student, interviewed all informants. One focus group interview with four participants was conducted face-to-face by the first author and one co-moderator present. The second focus group and individual interviews used a secure digital platform. All participants gave one interview. Notes were taken during and immediately after each interview. The interviews were conducted between November 2020 and March 2021.

The authors developed the interview guide for this study. The questions were based on previous literature (24), the experiences of young people with CFS/ME, and input from teachers experienced with young people with CFS/ME. The interview guide consisted of three main questions; "What, in your experience, is challenging for students with CFS/ME concerning the school?", "What experiences do you have from adapting education for these students?", and "How has the COVID-19 pandemic impacted the school day for students with CFS/ME?". The open-ended questions allowed the participants to speak freely about themes related to questions not yet asked. The first and last author edited the interview guide before the individual interviews based on data from the focus group interviews. This edition supplemented the interview guide with questions on how the school personnel, school nurses, and other professionals cooperated when planning education for students with CFS/ME. Interviews were audio-recorded and transcribed verbatim. The transcripts included only the participants occupation. The focus group interviews lasted 75–86 min, whereas the individual interviews lasted 61–82 min.

Data Analysis

The analyses were conducted with Malterud's Systematic Text Condensation (STC), a descriptive cross-case analysis strategy involving an iterative four-step analysis procedure developed from Giorgi's psychological phenomenological analysis (25). In step 1, reading all transcripts gave an overall impression of the data. In step 2, meaningful units in the transcripts were coded and grouped into five preliminary themes (**Table 2**). Step 3 included a systematic abstraction of the meaningful units by reducing the content into a condensate that maintained the informants' phrasings. In step 4, the first author wrote the analytical text and discussed it repeatedly with the co-authors, adding quotes to illustrate the findings. **Table 2** provides an illustration of the analyses. Preliminary results were presented and commented on in a national interdisciplinary forum experienced with young people with CFS/ME and discussed with a research group on patient education and participation at the university consisting of researchers experienced with qualitative methods.

The results were repeatedly checked against transcripts for validation. Identified themes and meaningful units were checked and recognized to verify that the final results were related to the original data. Microsoft Excel Version 2008 (26) and MindManager 2020 (27) were used as systematization tools during the analyses.

Ethical Considerations

All procedures performed were in accordance with and approved by the standards of The Norwegian Center for Research Data (approval number 420197) and performed according to the Declaration of Helsinki. Consent was obtained from municipal directors to recruit participants among the associated municipalities' employees. All informants received oral and written information about the study and gave informed and written consent to participate. Each participant had a number used during the focus group interviews to avoid the use of names. Each participant was also de-identified by using of numbers in transcripts. The names of schools or districts were not mentioned in the transcripts. Data were stored and handled according to government regulations and the regulations of the organization responsible for the project.

RESULTS

In total, 12 participants, 10 females and 2 males, were interviewed (**Table 1**). Six of them knew their students before they got CFS/ME, whereas six had met the student only after they fell ill. The participants talked about experiences on educational adaptations for students from both before and after the students received the CFS/ME diagnosis. They had little experience with educational adaptations for students with CFS/ME from the lockdown of schools in the COVID-19 pandemic. The participants' experiences were analyzed and categorized into the four themes; *Adaptation of education before the student is diagnosed is challenging, We experience that students lose*

TABLE 2 | Illustration of the four steps of systematic text condensation.

Step 1 Preliminary themes	Step 2 Meaningful units coded and grouped in five preliminary themes	Step 3 Condensed meaning (Preliminary themes were gathered in four main themes)	Step 4 Final themes (Themes were renamed, and quotes were added)
Distance relations expectation trust	**(1) Cognitive and emotional challenges in students with CFS/ME** "They fall outside while friends and peers move on," "Students have lost confidence in school." **(2) Relations with student and parents** "We communicate mostly with parents…," "We do not know when the student will attend school," "We are supposed to treat them as regular students"	**Lack of competence** We were supposed to treat the student as a regular student when he attended school, but this was often not possible because the student did not know what the class worked on. It has been challenging that the students were absent for an extended period and that we had little access to them.	**We experience that the students lose confidence in school**
Dilemma academically or socially	**(3) Adaptations of education** "The assessment is year-long," "Continuity of education is challenging when the student cannot meet regularly at school or are absent for such a long time," "It has been a lot of trying and failing"	**Demanding matters to be engaged in over time** The challenges lie in that the assessment period is often year-long. There are uncertainties around CFS/ME, and without knowledge, it has been difficult for us as teachers to make adequate adaptations for these students. It is necessary to focus on the social aspect as well as the academic. There has been a lot of trying and failing. Measures we have made often did not work.	**Adaptations of education before the student is diagnosed is challenging**
Affiliation	**(4) Social adaptations at school** "The contact with school is important, they need to feel that they take part in things that happen"		
Helplessness Communication	**(5) Interdisciplinary collaboration.** "It is a relief when diagnosis is set, then we are informed from the hospital," "Health care providers have a better awareness around CFS/ME now than previously," "Information from health care providers is clear"	**Important with interdisciplinary collaboration.** It has been a great relief when the students are diagnosed, and we receive information from health care providers. We have experienced that health care providers have more knowledge about CFS/ME now than previously and that it is essential with a steady plan for adaptations of education and social life at school.	**Interdisciplinary collaboration is valuable but sometimes challenging**
Understanding	"Perhaps we should have started earlier," "School absence is not problematized enough."	School absence is poorly problematized. We do not need a diagnosis to initiate measures.	**Suggestions on successful adaptations**

confidence in school, Interdisciplinary collaboration is valuable but sometimes challenging, and Suggestions on successful adaptations.

Adaptation of Education Before the Student Is Diagnosed Is Challenging

A perceived key challenge in educational adaptations for students with CFS/ME was the often year-long assessment process before the students received a diagnosis. This was a shared experience among all the participants. Teachers especially, spoke about educational adaptations. In this period, it was not necessarily straightforward for teachers to introduce educational adaptations because they lacked experience with CFS/ME and the necessary understanding of the student's needs. Therefore, they could easily make mistakes. However, if the teachers waited for a diagnosis before they initiated adaptive measures, students could be pressured to be more active than they could cope with and, thus, get worse. According to the school nurses, there were differences in whether teachers appropriately assessed the severity of the student's problems. Thus, early adaptations at school varied a lot. This could lead to unnecessary loss of education and social contact for the students, possibly followed by a lost sense of belonging to the school. In high school primarily, the challenges with adaptations were related to expectations of that students usually became more independent and made their own choices, while these students often became more dependent on others.

> "(…) I can honestly say that the biggest challenge I experience as a school nurse with this patient group is before they receive the diagnosis (…) the period from a loss of function until they receive

the diagnosis, it's so tough, the air flows out of the balloon." (*School nurse III*)

Teachers found it challenging to maintain or establish a relationship with the students because of their student's high levels of school absence and hence, limited contact with the students. They usually communicated with the parents to reduce the burden on the student. Due to fluctuating symptoms, the teachers often did not know whether the student was coming to school or not on a particular day. When the student attended school, it was challenging to re-connect the student with the classmates because he often did not know what the class worked on.

"(…) It is difficult for me to keep track of what the student has learned academically, (…), did you attend the previous class, no you did not, you attended that one, but not the one before, so keeping track is very challenging, I think." (*Teacher IV*)

We Experience That the Students Lose Confidence in School

A shared experience among the participants was that they had met students who had lost confidence in school because their struggles were not taken seriously by the school and because expectations toward them were too high. For instance, one of the participants talked about a student who had to run 6 km during the physical education class because that was the scheduled activity that day. Another example was that missed training on key concepts could lead to embarrassment in the classroom when students had to answer questions requiring knowledge about the taught key concepts. During the classes, the informants had this in mind because the loss of confidence and embarrassments among peers could lead to school refusal in the students.

"It is like both unknown and scary waters (…) so if we (…) push too hard, have too large expectations that lead to a burden in the form of performance pressure, but at the same time does not have too low expectations so that he has nothing to reach for or something to be proud of (…). He cannot do everything. He must be re-engaged in some way, but it has to work. It is a somewhat delicate balance." (*Teacher IV*)

The school nurses, especially talked about how their expertise in preventive health care could be helpful in this. By expressing needs on behalf of the students, they could contribute to a mutual understanding between students, parents, and teachers, thereby preventing the student's loss of confidence. In situations where teachers felt insecure, school nurses said they could provide support to reassure teachers about the adaptations they made for the students.

"It is not like we meet a lot of these students either, but for the individual teacher it will probably be the first time (…), so I think then we have more experience than them anyway (…)." (*School nurse IV*)

Even though the participants had experienced success with minor educational adaptations, the participants also experienced that the adaptations at school did not help much.

"(…) it becomes such a hopeless situation for a helper, we should not take over their problems, but there is great compassion for them, and that can be challenging (…). You have no prognosis or future, and it is so hurtful to deal with." (*School nurse IV*)

Interdisciplinary Collaboration Is Valuable but Sometimes Challenging

The participants experienced great relief when the students received a diagnosis. Subsequently, the students received follow-up from the hospital, and both teachers, counselors, and school nurses could dialogue with the hospital. For all participants, health care personnel had to communicate clearly to the school about the student's challenges. Participants, especially teachers with little experience, said they needed firsthand information to understand what adaptations could benefit students with CFS/ME.

"(…) I think the person who attends the collaborative meetings, like the teacher, will be the one who has the best understanding of the problem and the measures, (…) I feel I miss a bit because I do not attend these meetings with the medical staff and the EPS." (*Teacher II*)

All participants highly valued the collaboration with health care providers because they were assured that the student's health was well taken care of, and they were guided on what measures to initiate. One participant with previous and current experience with CFS/ME, said that hospital personnel now referred to more evidence-based knowledge on CFS/ME regarding the need to focus on the social aspects and learning. Collaboration regarding students with substantial school absences was necessary for the participants. Nevertheless, interdisciplinary collaboration meetings could also be challenging.

"'Between the devil and the deep blue sea' can be the feeling because the school can be frustrated and the parents can be frustrated, and they are often frustrated with each other, so that enabling a good collaboration is sometimes challenging, because the parents have been coping with it for years, and the school thinks quite traditionally" (*EPS II*)

Another challenge regarding interdisciplinary collaboration, especially talked about by teachers and counselors, was related to different theories about what causes CFS/ME. In interdisciplinary meetings, they could spend a long time and use much energy discussing this issue. The interdisciplinary collaboration was nevertheless a joint project where the school had a significant responsibility to show interest and make contact, and where parents were the connection between the student, school, and health care.

"(…) It is so vital how the school copes with this situation when the student becomes so ill, and then I think specifically about the importance of a teacher making contact once a week, even if there

TABLE 3 | Summary of challenges related to adaptation of education and social life at school for young people with CFS/ME.

Challenges before diagnosis	Students have lost confidence in school	Challenges in interdisciplinary collaboration
Lacked experience and understanding of the student's needs.	Important to know how much you can push the student.	Frustrations such as obstacles to collaboration.
Lacked contact with the student.	Adaptations did not help much.	Uncertainty about CFS/ME etiology.
Students could be pressured to do more than they could cope with.	School nurses can support teachers with early adaptations.	Exhausting meetings.

is no teaching (…). Just the fact that he came by and said hello and talked a little (…) it meant so much to the student, (…) it was the lifeline to society in a way, and that he could send greetings back to the classmates." (*School nurse III*)

The interdisciplinary meetings involved many professionals from both health care and educational systems. In addition, the attending professionals could change over time. The participants had experienced that the interdisciplinary meetings with many participants could lead to exhausted students and parents. Nevertheless, the interdisciplinary meetings were valuable, particularly if they agreed on a concrete plan with realistic educational adaptations.

"A concrete plan with specific goals (…) have their adapted plan where they in a way see what they are going to do, and to feel that they can attain the goal. Getting a confirmation that what they are doing is good enough and that it is what we expect (…) is important." (*Teacher V*)

A summary of the challenges school personnel and school nurses had experienced is provided in **Table 3**.

Suggestions on Successful Adaptations

The participants suggested ways for how successful adaptations could be carried out. For instance, counselors and school nurses pointed out that one measure could ensure that school absence was problematized early. Another suggestion was that the students were referred to Educational, Psychological Counseling Service (EPS) as early as possible. This presupposed that the school collaborated and trusted that EPS initiated the necessary measures in collaboration with the students and their parents. Teachers had experienced increased contact with students with CFS/ME by using digital teaching during the school lock down due to the COVID-19 pandemic. They suggested that this could be a continued measure beyond the pandemic if they had the resources to develop digital teaching for students with CFS/ME, for instance, as a tool for individual counseling time with the student.

"(...) measures we do for these students are to give work tasks that provide empowerment (…). Then I also feel that the student is motivated to do this again. (…) it may be to ask some leading questions or do something that they are a little familiar with, then, to make things safer (…) We have always placed the student with someone he knows well (…) I think it is a motivating factor to come to school as well." (*Teacher I*).

Because the participants, especially teachers, had only met a few students with CFS/ME, they needed more knowledge about suitable educational adaptations for these students. Related to this, they talked about the importance of collaboration between schools. One teacher said that now it seemed as if they were "reinventing the wheel" repeatedly in their respective schools. Through meeting students with CFS/ME, teachers gained experience and knowledge potentially beneficial for others. Therefore, they suggested it could be a good thing to have arenas where teachers could exchange experiences on educational adaptation for students with CFS/ME. They also said it could be helpful to meet students who had recovered from CFS/ME to hear their stories about what they considered to be beneficial adaptations at school. **Figure 1** provides the participants' suggestions for successful adaptations.

DISCUSSION

In this study, we explored teachers, counselors, and school nurses' experiences with making educational adaptations for students with CFS/ME. The main findings were that they experienced challenges with educational adaptations, especially before a CFS/ME diagnosis. The challenges were related to school absence, few opportunities to meet with the students, and uncertainty about the diagnosis and the students' adaptational needs. This impacted the teacher-student relationship, challenged a maintained continuity of the student's education, and the student could lose confidence in school. Suggestions for successful adaptations were; early problematization of school absence and interdisciplinary collaboration on a concrete plan for adaptive measures; to focus on the important teacher-student relationship; and increasing competence in schools by exchanging experiences between schools. This could prevent unnecessary educational and social losses and secure a maintained development in students with CFS/ME. Recent experiences with digital teaching during the COVID-19 pandemic gave knowledge possibly useful for the development of adaptive measures.

The Early Problematization of School Absence and Interdisciplinary Collaboration

Interdisciplinary collaboration was perceived as valuable when it provided necessary adaptive measures for students with CFS/ME. Continuity and well-defined plans for educational and social adaptations are of utmost importance for students with CFS/ME (1, 15). It also reassures teachers when they adapt education and relate to students with CFS/ME (1, 20). However, guidance on adaptive measures from specialized health care providers usually

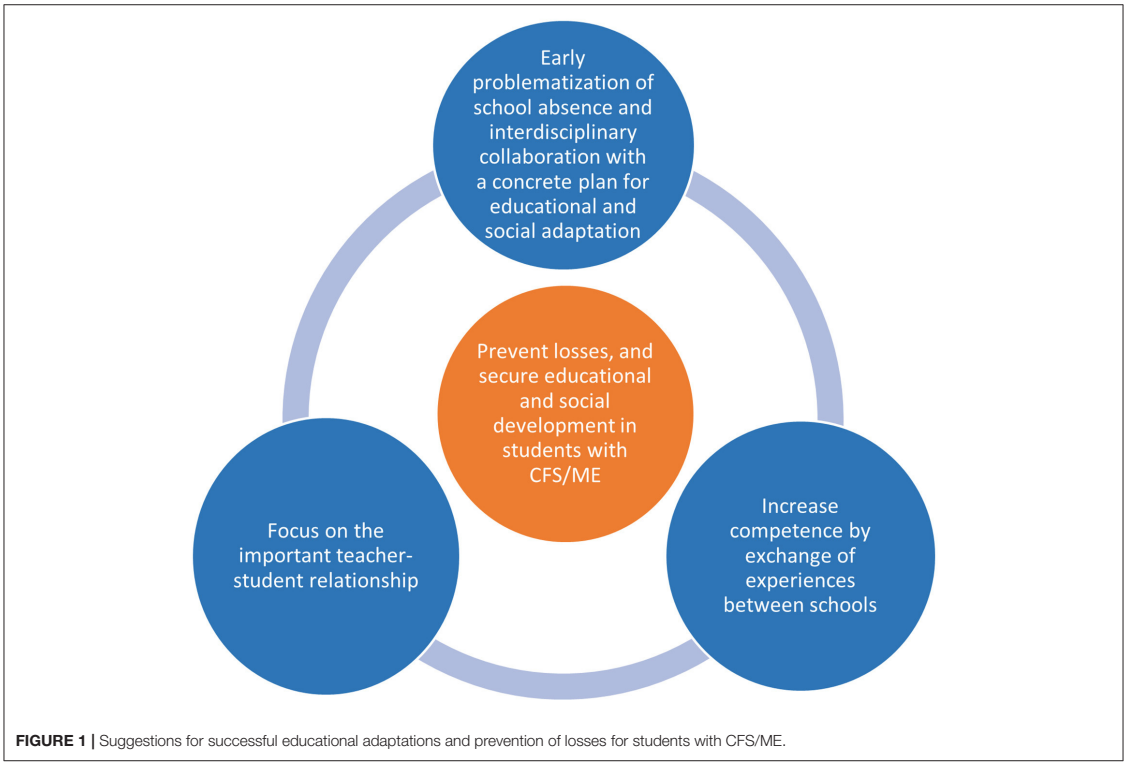

FIGURE 1 | Suggestions for successful educational adaptations and prevention of losses for students with CFS/ME.

comes after a diagnosis is given (4). It has been highlighted that early recognition and diagnosis of CFS/ME and persistent long-term interdisciplinary follow-up are important to reduce morbidity in young people with CFS/ME (28). The participants in the current study suggested that early problematization of school absence followed by early adaptive measures could prevent losses of academic and social skills among students with CFS/ME. The current study confirms that interdisciplinary collaboration, including health care providers, is valuable for educational adaptations for students with CFS/ME. In addition, the study adds that early assistance from teachers and experienced health care providers may prevent some of the academic and social losses among students with CFS/ME.

Teachers, counselors, and school nurses experienced several challenges related to educational adaptations for students with CFS/ME, especially before they received guidance from specialized health care providers. Recently, it was found that teachers' efforts to introduce early measures had been based on the teacher's educational expertise, conversations with the students' parents, and intuition, rather than knowledge about adaptive measures for students with CFS/ME (20). This led to variations in the kind of support the students were offered. Another study found that students with CFS/ME experienced not being believed about their challenges due to a lack of knowledge about CFS/ME in schools (29, 30). Uncertainties regarding the CFS/ME diagnosis and etiology and a wide specter of symptoms that frequently overlap with other conditions have made it difficult for students with CFS/ME to explain their illness to teachers and peers (29). Without a proper understanding or adaptive measures at school, students with CFS/ME experience school absence, poor school-related quality of life, and reduced academic performance (31). The loss of education and social life may lead to depressive thoughts and anxiety in students with CFS/ME (29, 30, 32). Thus, teachers, counselors, and school nurses need education about how CFS/ME impacts academic performance and social life in affected students (1, 20, 33).

Focus on the Essential Teacher-Student Relationship

Teachers experienced that it was challenging to maintain or initiate a teacher-student relationship, mostly due to the student's school absence. Thus, it was challenging to ensure continuity of the student's education. School attendance for students with CFS/ME may vary over time, and teachers often communicate with the parents instead of the student to minimize the pressure on the student (14, 20). It was previously found that students with CFS/ME who could not attend school struggled to be seen by their surroundings (34). Furthermore, increased contact with teachers was found to be associated with higher levels of health-related quality of life (HRQoL) in young people with CFS/ME (35). An important finding in this study is that teachers sometimes found it challenging to establish or

maintain a relationship with students with CFS/ME, and that this may impact continuity of education. A closer teacher-student relationship might facilitate the teacher's understanding of the students' individual needs and ability to provide support (20). In addition, this may also be a possibility for bedbound students to contact the school society and receive help with education (18). Digital teaching may improve the teacher-student contact for students with CFS/ME and improve the understanding of social isolation among young people with CFS/ME (36). Therefore, it may be helpful to learn from recent experiences related to social isolation, digital teaching, and contact with students during the COVID-19 pandemic (37).

Increase Competence in Schools by Exchange of Knowledge Between Schools

There was a limited exchange of knowledge between schools regarding adaptive measures for students with CFS/ME. A recent study describing the basis for teachers ' early adaptation of education for students with CFS/ME, did not mention the exchange of experiences between schools as a resource (20). Nevertheless, teacher collaboration and knowledge sharing can provide collegial support and improve educational adaptations for this group of students (38, 39), and this study supports that exchange between schools regarding successful adaptations of education for students with CFS/ME is a potential resource that is unexploited.

Implications

Interdisciplinary collaboration around students with CFS/ME should be initiated earlier in the course of the disease, even before diagnosis. By problematizing school absences early, students with CFS/ME symptoms can be identified and receive adaptive measures to prevent unnecessary losses pertaining to their education. It is also important to focus on maintaining or establishing a relationship between teachers and students with CFS/ME. Digital contact may be a valuable resource for this. Exchange of experiences between schools about early adaptive measures for students with CFS/ME is a resource with potential benefits for teachers when they adapt education for students with CFS/ME.

Strengths and Limitations

A strength of the study was the variation of experiences among the participants who represented three different professionality's in seven different schools. It was also a strength that the participants talked about both positive and negative experiences with the educational adaptations for students with CFS/ME. For reflexivity, preliminary results were discussed with others during the analyses.

A limitation to the study is that the sample was relatively small and limited to participants in Mid- Norway. If more districts and other countries were included, and the data collection period had been expanded, more participants could have been recruited, and more experiences could have been obtained.

The transfer from focus group to individual interviews made it possible to explore more expansive preliminary findings from the focus group interviews. Nevertheless, a limitation was that this reduced the possibility of obtaining data from the group dynamic in a focus group interview. Another limitation was that only a few participants represented high schools. Nevertheless, the findings include factors relevant both for secondary and high schools. A possible bias was that only participants with specific experiences registered. However, the various findings show that this was not the case.

It was a strength that a co-moderator participated in the first focus group interview, and a limitation that only one researcher conducted the digital interviews. However, the interviews were repeatedly discussed among the co-authors for input.

CONCLUSION

Teachers, counselors, and school nurses found it challenging to adapt education for students with CFS/ME. If school absence is problematized early, and if teachers, counselors, and school nurses get early assistance from experienced health care professionals, it may be possible to identify students with CFS/ME symptoms and initiate early adaptive measures. Interdisciplinary focus on a clear and constructive common plan with adaptive measures including maintained teacher-student contact, and educational and social adaptations may prevent the losses among students with CFS/ME. Recent experiences with digital teaching during the COVID-19 pandemic may provide proper adaptive measures for use beyond the pandemic.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

AUTHOR CONTRIBUTIONS

WS collected, analyzed, and interpreted the data and was the main author of the manuscript. TN supervised the data collection, analyses and interpretation of data, and contributed to the writing of the manuscript. TR supervised the project, analyses and interpretation of data, and contributed to the writing of the manuscript. All authors read and approved the final findings and the final manuscript.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Appendix

1. Approvalsfrom the Regional Ethical Committee for Medical and Health Profession Research and the Norwegian Centre of Research Data – and request letters for the county and municipalities directors

2. Study information with consent to participate; for Studies 1 & 2, and for Study 3

3. Data collection tools. List of Questionnaires, additional questions and data collection tool for data from medical records for Study 1, and interviewguides for Studies 2 and 3

Appendix 1

Approvals from the Regional Ethical Committee for Medical and Health Profession Research and the Norwegian Centre of Research Data, and request letters for the county and municipalities directors.

REK
REGIONALE KOMITEER FOR MEDISINSK OG HELSEFAGLIG FORSKNINGSETIKK

Region:	Saksbehandler:	Telefon:	Vår dato:	Vår referanse:
REK sør-øst	Tove Irene Klokk	22845522	22.06.2017	2017/749/REK sør-øst A
			Deres dato:	**Deres referanse:**
			16.06.2017	

Vår referanse må oppgis ved alle henvendelser

Torstein Baade Rø
Barne- og Ungdomsklinikken

2017/749 Helse og helse-relatert livskvalitet hos ungdom med kronisk utmattelse

Forskningsansvarlig: St. Olavs Hospital HF, Oslo universitetssykehus HF
Prosjektleder: Torstein Baade Rø

Vi viser til tilbakemelding i ovennevnte forskningsprosjekt. Tilbakemeldingen ble behandlet av leder REK sør-øst A på fullmakt.

Prosjektbeskrivelse (revidert av REK)

Formålet med prosjektet er å undersøke hvordan det går med ungdom som har kronisk utmattelsessyndrom (CFS/ME) eller andre tilstander hvor utmattelse er hovedproblemet.

Mange av disse pasientene får liten oppfølging fra helsevesenet. Gjennom å kartlegge hva slags oppfølging disse ungdommene har fått fra helsevesen, skole og for øvrig ønsker forskerne å få ny kunnskap om hvordan pasientgruppen har det helsemessig og om hvilke tiltak som har best effekt. Dette kan i neste omgang føre til forbedring av rehabiliterings/behandlingstilbudet til pasientgruppen.

Studien inkluderer medisinske opplysninger som diagnose, symptomer, skolefravær og ledsagende sykdommer ved tidspunkt for utredning ved Barne- og ungdomsklinikken og i ettertid.

For den kvantitative delen av studien rekrutteres 75-100 deltakere blant ungdom som har vært til utredelse for kronisk utmattelse i perioden 2013-2016. Disse skal besvare spørreskjema om symptomer og livskvalitet, og også bli intervjuet. I tillegg skal det innhentes opplysninger om utredningen fra deltakernes pasientjournal.

For den kvalitative delen av studien (intervju) gjøres fortløpende vurdering av deltakelse basert på om ny informasjon oppnås. Sannsynligvis vil denne delen av studien kreve 10-20 deltakere.

Studien inkluderer barn i alderen 12 – 18 år. Begge foreldre skal samtykke på vegne av barn under 16 år, mens ungdom mellom 16-18 år samtykker på egne vegne.

Saksgang

Søknad om forhåndsgodkjenning ble behandlet av komiteen i møte 04.05.2017. Det ble besluttet å utsette vedtak i saken. Følgende inngikk i komiteens vurdering jf. brev av 19.05.2017.

«Komiteen synes dette er en viktig studie for en sykdomsgruppe som opplever å få liten oppfølging fra helsevesenet.

Enkelte av spørreskjemaene som skal benyttes finnes kun på svensk eller engelsk. Disse må oversettes til norsk og sendes til REK for godkjenning. Prosjektleder bes også vurdere om prosjektperioden skal utvides. Prosjektperioden omfatter, i tillegg til praktisk gjennomføring av studien, også forskning og publisering av de opplysninger som er innhentet. Det er en vanlig misforståelse at sammenstilling av data og publisering skal skje etter prosjektperiodens utløp og ikke innenfor perioden. Etter prosjektslutt skal altså dataene oppbevares, men ikke forskes på.»

Prosjektleder har sendt tilbakemelding mottatt 16.06.2017. Spørreskjema oversatt til norsk og revidert protokoll og intervjuguide er vedlagt tilbakemeldingen. I tilbakemeldingen inngikk følgende:

« 1. Vi vedlegger nå norsk versjon av spørreskjemaene EQ-5D (tidligere innsendt på svensk) og dePaul Questionnaire (tidligere innsendt på engelsk). Skjemaet CHDU9D har vi valgt å ta ut av studien da det vil kreve for mye ressurser å gjøre en god oversettelse, og protokollen er oppdatert i forhold til at CHUD9D er tatt ut.

2. Vi ønsker å utvide prosjektperiode til 31.12.2020 ihht komiteens anbefaling.

3. Det er gjort mindre endringer i layout på intervjuskjemaet for å lette kvantitativ analyse av noen av svarene. Det er ikke endret på ordlyd eller antall spørsmål, men revidert intervjuguide vedlegges.»

Ny vurdering

Tilbakemeldingen er vurdert av komiteens leder på delegert fullmakt og anses som tilfredsstillende i henhold til komiteens merknader.

Vedtak

Prosjektet godkjennes med hjemmel i helseforskningsloven §§ 9 og 33.

Godkjenningen er gitt under forutsetning av at prosjektet gjennomføres slik det er beskrevet i søknaden, protokollen og tilbakemeldingen, og de bestemmelser som følger av helseforskningsloven med forskrifter.

Godkjenningen gjelder til 31.12.2020.

Av dokumentasjonshensyn skal opplysningene oppbevares i 5 år etter prosjektslutt. Opplysningene skal oppbevares avidentifisert, dvs. atskilt i en nøkkel- og en datafil. Opplysningene skal deretter slettes eller anonymiseres.

Forskningsprosjektets data skal oppbevares forsvarlig, se personopplysningsforskriften kapittel 2, og Helsedirektoratets veileder for «Personvern og informasjonssikkerhet i forskningsprosjekter innenfor helse- og omsorgssektoren».

Dersom det skal gjøres endringer i prosjektet i forhold til de opplysninger som er gitt i søknaden, må prosjektleder sende endringsmelding til REK, jf. helseforskningsloven § 11.

Prosjektet skal sende sluttmelding på eget skjema, jf. helseforskningsloven § 12, senest et halvt år etter prosjektslutt.

Klageadgang

Komiteens vedtak kan påklages til Den nasjonale forskningsetiske komité for medisin og helsefag, jf. helseforskningsloven § 10 tredje ledd og forvaltningsloven § 28. En eventuell klage sendes til REK sør-øst A. Klagefristen er tre uker fra mottak av dette brevet, jf. forvaltningsloven § 29.

Med vennlig hilsen

Knut Engedal
Professor dr. med.
Leder

Tove Irene Klokk
Rådgiver

Kopi til: elisabeth.selvaag@stolav.no, terje.rootwelt@ous.no, Oslo universitetssykehus HF ved øverste administrative ledelse: oushfdlgodkjenning@ous-hf.no, St. Olavs Hospital ved øverste administrative ledelse: post.adm.dir@stolav.no

REK
REGIONALE KOMITEER FOR MEDISINSK OG HELSEFAGLIG FORSKNINGSETIKK

Region:	Saksbehandler:	Telefon:	Vår dato:	Vår referanse:
REK sør-øst A	Tove Irene Klokk	22845522	01.07.2020	8459
			Deres referanse:	

Torstein Baade Rø

8459 Helse og helse-relatert livskvalitet hos ungdom med kronisk utmattelse

Forskningsansvarlig: St. Olavs Hospital HF

Søker: Torstein Baade Rø

REKs vurdering

Vi viser til søknad om prosjektendring datert 12.06.2020 for ovennevnte forskningsprosjekt. Søknaden er behandlet av leder REK sør-øst A på fullmakt, med hjemmel i helseforskningsloven § 11.

Det er ønskelig med et oppfølgingsspørsmål pr. telefon til de 20 deltakerne som har deltatt i den kvalitative delen av forskningsprosjektet. Spørsmålet gjelder hvordan ungdommene har opplevd tilrettelagt undervisning under Covid-19 perioden, og om det har hatt betydning for kontakten med skole. Protokollen er oppdatert med denne endringen.

Komiteens leder har vurdert endringen og har ingen innvendinger mot at denne gjennomføres slik som beskrevet.

Vedtak

Godkjent

Komiteen godkjenner med hjemmel i helseforskningsloven § 11 annet ledd at prosjektet videreføres i samsvar med det som fremgår av søknaden om prosjektendring, og i samsvar med de bestemmelser som følger av helseforskningsloven med forskrifter.

Vi gjør samtidig oppmerksom på at etter ny personopplysningslov må det også foreligge et behandlingsgrunnlag etter personvernforordningen. Det må forankres i egen institusjon.

Godkjenningen gjelder til 31.12.2020.

Dersom det skal gjøres ytterligere endringer i prosjektet i forhold til de opplysninger som er gitt i søknaden, må prosjektleder sende ny endringsmelding til REK.

Av dokumentasjonshensyn skal opplysningene oppbevares i 5 år etter prosjektslutt.

Opplysningene skal oppbevares avidentifisert, dvs. atskilt i en nøkkel- og en datafil. Opplysningene skal deretter slettes eller anonymiseres.

Prosjektet skal sende sluttmelding til REK, se helseforskningsloven § 12, senest 6 måneder etter at prosjektet er avsluttet.

Vennlig hilsen

Knut Engedal
Professor dr. med.
Leder REK sør-øst A

Tove Irene Klokk
Seniorrådgiver
REK sør-øst

Kopi til forskningsansvarlig institusjon(er) og medbruker(e).

Klageadgang
Du kan klage på komiteens vedtak, jf. forvaltningsloven § 28 flg. Klagen sendes til REK sør-øst A. Klagefristen er tre uker fra du mottar dette brevet. Dersom vedtaket opprettholdes av REK sør-øst A, sendes klagen videre til Den nasjonale forskningsetiske komité for medisin og helsefag (NEM) for endelig vurdering.

REK
REGIONALE KOMITEER FOR MEDISINSK OG HELSEFAGLIG FORSKNINGSETIKK

Region:	Saksbehandler:	Telefon:	Vår dato:	Vår referanse:
REK sør-øst A	Tove Irene Klokk	22845522	07.10.2020	8459
			Deres referanse:	

Torstein Baade Rø

8459 Helse og helse-relatert livskvalitet hos ungdom med kronisk utmattelse

Forskningsansvarlig: St. Olavs Hospital HF

Søker: Torstein Baade Rø

REKs vurdering

Vi viser til søknad om prosjektendring datert 05.10.2020 for ovennevnte forskningsprosjekt (REK 2017/749). Søknaden er behandlet av sekretariatet i REK sør-øst på delegert fullmakt fra REK sør-øst A, med hjemmel i helseforskningsloven § 11.

Det søkes om å inkludere to nye medarbeidere i prosjektet:

- Torunn Hatlen Nøst, førsteamanuensis, NTNU.
- Liv Solvår Nymark, forsker, St. Olavs Hospital HF.

Sekretariatet har vurdert endringene og har ingen innvendinger mot at disse gjennomføres.

Vedtak

Godkjent

Komiteen godkjenner med hjemmel i helseforskningsloven § 11 annet ledd at prosjektet videreføres i samsvar med det som fremgår av søknaden om prosjektendring i samsvar med de bestemmelser som følger av helseforskningsloven med forskrifter.

Vi gjør samtidig oppmerksom på at etter ny personopplysningslov må det også foreligge et behandlingsgrunnlag etter personvernforordningen. Det må forankres i egen institusjon.

Godkjenningen gjelder til 31.12.2020.

Dersom det skal gjøres ytterligere endringer i prosjektet i forhold til de opplysninger som er gitt i søknaden, må prosjektleder sende ny endringsmelding til REK.

Av dokumentasjonshensyn skal opplysningene oppbevares i 5 år etter prosjektslutt. Opplysningene skal oppbevares avidentifisert, dvs. atskilt i en nøkkel- og en datafil.

Opplysningene skal deretter slettes eller anonymiseres.

Prosjektet skal sende sluttmelding til REK, se helseforskningsloven § 12, senest 6 måneder etter at prosjektet er avsluttet.

Vennlig hilsen

Jacob C. Hølen
Sekretariatsleder
REK sør-øst

Tove Irene Klokk
Seniorrådgiver
REK sør-øst

Kopi til forskningsansvarlig institusjon(er) og medbruker(e).

Klageadgang
Du kan klage på komiteens vedtak, jf. forvaltningsloven § 28 flg. Klagen sendes til REK sør-øst A. Klagefristen er tre uker fra du mottar dette brevet. Dersom vedtaket opprettholdes av REK sør-øst A, sendes klagen videre til Den nasjonale forskningsetiske komité for medisin og helsefag (NEM) for endelig vurdering.

REK
REGIONALE KOMITEER FOR MEDISINSK OG HELSEFAGLIG FORSKNINGSETIKK

Region:	Saksbehandler:	Telefon:	Vår dato:	Vår referanse:
REK sør-øst A	Tove Irene Klokk	22845522	02.11.2020	8459
			Deres referanse:	

Torstein Baade Rø

8459 Helse og helse-relatert livskvalitet hos ungdom med kronisk utmattelse

Forskningsansvarlig: St. Olavs Hospital HF

Søker: Torstein Baade Rø

REKs vurdering

Vi viser til søknad om prosjektendring datert 28.10.2020 for ovennevnte forskningsprosjekt (REK 2017/749). Søknaden er behandlet av sekretariatet i REK sør-øst på delegert fullmakt fra REK sør-øst A, med hjemmel i helseforskningsloven § 11.

Det søkes om å utvide prosjektperioden, med ny sluttdato 31.12.2021.

Datainnsamlingen i prosjektet er avsluttet i henhold til plan, men det er behov for mer tid for å ferdigstille analyser og publikasjoner.

Sekretariatet har vurdert endringen og har ingen innvendinger mot at denne gjennomføres.

Vedtak

Godkjent

Komiteen godkjenner med hjemmel i helseforskningsloven § 11 annet ledd at prosjektet videreføres i samsvar med det som fremgår av søknaden om prosjektendring i samsvar med de bestemmelser som følger av helseforskningsloven med forskrifter.

Vi gjør samtidig oppmerksom på at etter ny personopplysningslov må det også foreligge et behandlingsgrunnlag etter personvernforordningen. Det må forankres i egen institusjon.

Godkjenningen gjelder til 31.12.2021.

Dersom det skal gjøres ytterligere endringer i prosjektet i forhold til de opplysninger som er gitt i søknaden, må prosjektleder sende ny endringsmelding til REK.

Av dokumentasjonshensyn skal opplysningene oppbevares i 5 år etter prosjektslutt. Opplysningene skal oppbevares avidentifisert, dvs. atskilt i en nøkkel- og en datafil.

Opplysningene skal deretter slettes eller anonymiseres.

Prosjektet skal sende sluttmelding til REK, se helseforskningsloven § 12, senest 6 måneder etter at prosjektet er avsluttet.

Vennlig hilsen

Jacob C. Hølen
Sekretariatsleder
REK sør-øst

Tove Irene Klokk
Seniorrådgiver
REK sør-øst

Kopi til forskningsansvarlig institusjon(er) og medbruker(e).

Klageadgang
Du kan klage på komiteens vedtak, jf. forvaltningsloven § 28 flg. Klagen sendes til REK sør-øst A. Klagefristen er tre uker fra du mottar dette brevet. Dersom vedtaket opprettholdes av REK sør-øst A, sendes klagen videre til Den nasjonale forskningsetiske komité for medisin og helsefag (NEM) for endelig vurdering.

Godkjenning fra Norsk senter for forskningsdata 14.07.2020

Det innsendte meldeskjemaet med referansekode 420197 er nå vurdert av NSD. Følgende vurdering er gitt: Det er vår vurdering at behandlingen av personopplysninger i prosjektet vil være i samsvar med personvernlovgivningen så fremt den gjennomføres i tråd med det som er dokumentert i meldeskjemaet 14.07.2020 med vedlegg, samt i meldingsdialogen mellom innmelder og NSD. Behandlingen kan starte.

MELD VESENTLIGE ENDRINGER Dersom det skjer vesentlige endringer i behandlingen av personopplysninger, kan det være nødvendig å melde dette til NSD ved å oppdatere meldeskjemaet. Før du melder inn en endring, oppfordrer vi deg til å lese om hvilke type endringer det er nødvendig å melde: nsd.no/personvernombud/meld_prosjekt/meld_endringer.html Du må vente på svar fra NSD før endringen gjennomføres.

TYPE OPPLYSNINGER OG VARIGHET: Prosjektet vil behandle alminnelige kategorier av personopplysninger frem til 31.12.2021. LOVLIG GRUNNLAG: Prosjektet vil innhente samtykke fra de registrerte til behandlingen av personopplysninger. Vår vurdering er at prosjektet legger opp til et samtykke i samsvar med kravene i art. 4 og 7, ved at det er en frivillig, spesifikk, informert og utvetydig bekreftelse som kan dokumenteres, og som den registrerte kan trekke tilbake. Lovlig grunnlag for behandlingen vil dermed være den registrertes samtykke, jf. personvernforordningen art. 6 nr. 1 bokstav a.

PERSONVERNPRINSIPPER: NSD vurderer at den planlagte behandlingen av personopplysninger vil følge prinsippene i personvernforordningen om: - lovlighet, rettferdighet og åpenhet (art. 5.1 a), ved at de registrerte får tilfredsstillende informasjon om og samtykker til behandlingen - formålsbegrensning (art. 5.1 b), ved at personopplysninger samles inn for spesifikke, uttrykkelig angitte og berettigede formål, og ikke viderebehandles til nye uforenlige formål - dataminimering (art. 5.1 c), ved at det kun behandles opplysninger som er adekvate, relevante og nødvendige for formålet med prosjektet - lagringsbegrensning (art. 5.1 e), ved at personopplysningene ikke lagres lengre enn nødvendig for å oppfylle formålet

DE REGISTRERTES RETTIGHETER: Så lenge de registrerte kan identifiseres i datamaterialet vil de ha følgende rettigheter: åpenhet (art. 12), informasjon (art. 13), innsyn (art. 15), retting (art. 16), sletting (art. 17), begrensning (art. 18), underretning (art. 19), dataportabilitet (art. 20). NSD vurderer at informasjonen som de registrerte vil motta oppfyller lovens krav til form og innhold, jf. art. 12.1 og art. 13. Vi minner om at hvis en registrert tar kontakt om sine rettigheter, har behandlingsansvarlig institusjon plikt til å svare innen en måned.

FØLG DIN INSTITUSJONS RETNINGSLINJER: NSD legger til grunn at behandlingen oppfyller kravene i personvernforordningen om riktighet (art. 5.1 d), integritet og konfidensialitet (art. 5.1. f) og sikkerhet (art. 32). For å forsikre dere om at kravene oppfylles, må dere følge interne retningslinjer og eventuelt rådføre dere med behandlingsansvarlig institusjon. OPPFØLGING AV PROSJEKTET NSD vil følge opp underveis (hvert annet år) og ved planlagt avslutning for å avklare om behandlingen av personopplysningene pågår i tråd med den behandlingen som er dokumentert. Lykke til med prosjektet!

Kontaktperson hos NSD: Henrik Netland Svensen

NTNU
Kunnskap for en bedre verden

Til Fylkesdirektør for utdanning i Trøndelag Fylkeskommune Trondheim, 24.8.20

Forespørsel om godkjenning av forskningsprosjekt blant lærere og rådgivere i videregående skole

Ved NTNU arbeider vi med et ph.d. prosjekt som undersøker livskvalitet og skolehelse blant ungdom med kronisk utmattelsessyndrom. Vi har tidligere undersøkt ungdommenes livskvalitet og funnet at skolen og kontakten med lærere kan være assosiert med høyere livskvalitet. På bakgrunn av dette ønsker vi å undersøke hvilke erfaringer lærere, rådgivere og helsesykepleiere har fra arbeid med å legge til rette for en god skolehverdag for ungdom med kronisk utmattelsessyndrom. Hensikten er også å få fram data som kan være nyttige i arbeidet med Fagfornyelsesplanen og det tverrfaglige livsmestringsfaget for elevene.

Studien ønsker å rekruttere lærere, rådgivere og helsesykepleiere til å delta i et fokusgruppeintervju med en varighet på 2 timer, en gang per deltager. Det planlegges å gjennomføre 2-4 fokusgruppeintervju med 6-8 deltagere i hver gruppe. Det er ønskelig å gjennomføre fokusgruppeintervjuene i løpet av høsten 2020. Hvis koronavirussituasjonen tilsier det vil digital fokusgruppeundersøkelse benyttes. Funnene vil formidles i publikasjoner og på seminarer.

NSD har vurdert og gitt tilrådning til prosjektet (referanse 420197).

Svar på vår forespørsel og eventuelle spørsmål kan sendes til undertegnede på e-post:

Med vennlig hilsen,

Wenche Ann Similä,
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Telefon: 73 41 25 34

Vedlegg: Informasjonsskriv og samtykkeskjema til deltagere i studien

NTNU
Kunnskap for en bedre verden

Til Kommunedirektøren i …….. kommune, Trondheim, 31.8.20

Forespørsel om godkjenning av forskningsprosjekt blant lærere, rådgivere og helsesykepleiere i ungdomsskolen

Ved NTNU arbeider vi med et ph.d. prosjekt som undersøker livskvalitet og skolehelse for ungdom med kronisk utmattelsessyndrom. Vi har tidligere undersøkt ungdommenes livskvalitet og funnet at skolen og kontakten med lærere kan være assosiert med høyere livskvalitet. På bakgrunn av dette ønsker vi å undersøke hvilke erfaringer lærere, rådgivere og helsesykepleiere har fra arbeid med å legge til rette for en god skolehverdag for ungdom med kronisk utmattelsessyndrom. Hensikten er også å få fram data som kan være nyttige i arbeidet med Fagfornyelsesplanen og det tverrfaglige livsmestringsfaget for elevene.

Studien ønsker å rekruttere lærere, rådgivere og helsesykepleiere til å delta i et fokusgruppeintervju med en varighet på 2 timer, en gang per deltager. Det planlegges å gjennomføre 2-4 fokusgruppeintervju med 6-8 deltagere i hver gruppe. Hvis koronavirussituasjonen tilsier det vil digital fokusgruppeundersøkelse benyttes. Det er ønskelig å gjennomføre fokusgruppeintervjuene i løpet av høsten 2020. Funnene vil formidles i publikasjoner og på seminarer for fagpersoner innen skole og helse.

NSD har vurdert og gitt tilrådning til prosjektet (referanse 420197).

Svar på vår forespørsel og eventuelle spørsmål kan sendes til undertegnede på e-post:

Med vennlig hilsen,

Wenche Ann Similä,
Ph.d. student
NTNU, Institutt for klinisk og molekylærmedisin.
wenche.a.simila@ntnu.no
Telefon: 72 82 56 27/99 40 18 30

Ansvarlig prosjektleder
Torstein Baade Rø,
Instituttleder, NTNU Institutt for klinisk og molekylærmedisin, MD, Phd.
torstein.ro@ntnu.no
Mobil: 99 61 40 25

Ansvarlig biveileder
Torunn Hatlen Nøst,
Førsteamanuensis,
NTNU, Institutt for psykisk helse
torunn.h.nost@ntnu.no
Telefon: 73 41 25 34

Vedlegg: Informasjonsskriv og samtykkeskjema til deltagere i studien

Appendix 2

Study information with consent to participate; for Studies 1 & 2, and for Study 3

ST. OLAVS HOSPITAL
UNIVERSITETSSYKEHUSET I TRONDHEIM

Oslo universitetssykehus

FORESPØRSEL OM DELTAKELSE I FORSKNINGSPROSJEKTET

HELSE OG LIVSKVALITET HOS UNGDOM MED KRONISK UTMATTELSE

Dette er et spørsmål til deg om å delta i et forskningsprosjekt for å undersøke helse og livskvalitet hos ungdom med kronisk utmattelse. Du får denne henvendelsen fordi du i løpet av de siste 4 årene har vært til utredning for langvarig utmattelse ved Barne- og ungdomsklinikken, St. Olavs Hospital eller Oslo Universitetssykehus.

HVA INNEBÆRER PROSJEKTET?

Ved Barne- og ungdomsklinikken har vi et utredningstilbud for barn og ungdom med kronisk utmattelse. Vi vet imidlertid for lite om hvordan det går etter utredningen hos oss med helseplager og livskvalitet, og målet med dette prosjektet er å finne ut hvordan du har hatt det etter utredningen hos oss. Dette kan identifisere tiltak som fungerer godt eller svakheter i helsetilbudet som vi i neste omgang kan gjøre noe med og forbedre for fremtidige pasienter.

Deltakelse i prosjektet medfører at du må svare på et spørreskjema om hvordan du har det (livskvalitet) og din helse. Dette vil ta 45-60 minutter av din tid. Videre ønsker vi også å intervjue deg på telefon eller på nærmere avtalt sted, dette vil ta ytterligere inntil 60 minutter. I tillegg henter vi opplysninger om utredningen hos oss fra din pasientjournal.

MULIGE FORDELER OG ULEMPER

Fordelen med deltakelse er at resultatene forhåpentligvis fører til en forbedring av vårt og andres tilbud til ungdom med langvarig utmattelse. Dette vil komme fremtidige pasienter til gode. Du vil få tilbud om en samtale med sykepleier etter intervjuet hvor du kan få råd vedrørende din videre oppfølging. Ulempene ved å delta er at det vil ta tid for deg å svare på spørreskjema og delta i intervjuet. Vi vil legge til rette for at intervjuet kan skje på en måte og et tidspunkt som passer deg slik at du ikke behøver å ta fri fra skole eller jobb.

FRIVILLIG DELTAKELSE OG MULIGHET FOR Å TREKKE SITT SAMTYKKE

Det er frivillig å delta i prosjektet. Dersom du ønsker å delta, tar du kontakt med Wenche Ann Similä (se kontaktinformasjon under) og undertegner samtykkeerklæringen på siste side. Du kan når som helst og uten å oppgi noen grunn trekke ditt samtykke. Dette vil ikke få konsekvenser for din videre behandling. Dersom du trekker deg fra prosjektet, kan du kreve å få slettet innsamlede prøver og opplysninger, med mindre opplysningene allerede er inngått i analyser eller brukt i vitenskapelige publikasjoner. Dersom du senere ønsker å trekke deg eller har spørsmål til prosjektet, kan du kontakte sykepleier, M.Sc. Wenche Ann Similä, epost wenche.ann.similae@stolav.no, telefon 72 82 56 27 eller prosjektleder og barnelege Torstein Baade Rø, torstein.ro@ntnu.no, telefon 72 57 40 51.

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Oslo universitetssykehus

HVA SKJER MED INFORMASJONEN OM DEG?

Informasjonen som registreres om deg skal kun brukes slik som beskrevet i hensikten med studien. Du har rett til innsyn i hvilke opplysninger som er registrert om deg og rett til å få korrigert eventuelle feil i de opplysningene som er registrert.

Alle opplysningene vil bli behandlet uten navn og fødselsnummer eller andre direkte gjenkjennende opplysninger. En kode knytter deg til dine opplysninger gjennom en navneliste.

Prosjektleder har ansvar for den daglige driften av forskningsprosjektet og at opplysninger om deg blir behandlet på en sikker måte. Informasjon om deg vil bli anonymisert eller slettet senest fem år etter prosjektslutt.

GODKJENNING

Prosjektet er godkjent av Regional komite for medisinsk og helsefaglig forskningsetikk, REK 2017/xx.

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UNIVERSITETSSYKEHUSET I TRONDHEIM

Oslo universitetssykehus

SAMTYKKE TIL DELTAKELSE I PROSJEKTET

JEG ER VILLIG TIL Å DELTA I PROSJEKTET

Sted og dato

Deltakers signatur

Deltakers navn med trykte bokstaver

Jeg bekrefter å ha gitt informasjon om prosjektet

Sted og dato

Signatur

Rolle i prosjektet

FORESPØRSEL OM DELTAKELSE I FORSKNINGSPROSJEKTET

HELSE OG LIVSKVALITET HOS UNGDOM MED KRONISK UTMATTELSE

Dette er et spørsmål til deg om å la ditt barn delta i et forskningsprosjekt for å undersøke helse og livskvalitet hos ungdom med kronisk utmattelse. Du får denne henvendelsen fordi barnet ditt i løpet av de siste 4 årene har vært til utredning for langvarig utmattelse ved Barne- og ungdomsklinikken, St. Olavs Hospital eller Oslo Universitetssykehus.

HVA INNEBÆRER PROSJEKTET?

Ved Barne- og ungdomsklinikken har vi et utredningstilbud for barn og ungdom med kronisk utmattelse. Vi vet imidlertid for lite om hvordan det går etter utredningen hos oss med helseplager og livskvalitet, og målet med dette prosjektet er å finne ut hvordan barnet ditt har hatt det etter utredningen hos oss. Dette kan identifisere tiltak som fungerer godt eller svakheter i helsetilbudet som vi i neste omgang kan gjøre noe med og forbedre for fremtidige pasienter.

Deltakelse i prosjektet medfører at du og ditt barn må svare på et spørreskjema om hvordan barnet har det helsemessig og mer generelt. Dette vil ta 45-60 minutter av deres tid. Videre ønsker vi også å intervjue deg og ditt barn på nærmere avtalt sted, dette vil ta ytterligere inntil 60 minutter. I tillegg henter vi opplysninger om utredningen hos oss fra barnets pasientjournal.

MULIGE FORDELER OG ULEMPER

Fordelen med deltakelse er at resultatene forhåpentligvis fører til en forbedring av vårt og andres tilbud til ungdom med langvarig utmattelse. Dette vil komme fremtidige pasienter til gode. Dere vil få tilbud om en samtale med sykepleier etter intervjuet hvor dere kan få råd vedrørende videre oppfølging. Ulempene ved å delta er at det vil ta tid for deg og barnet ditt å svare på spørreskjema og delta i intervjuet. Vi vil legge til rette for at intervjuet kan skje på en måte og et tidspunkt som passer dere slik at dere ikke behøver å ta fri fra skole eller jobb.

FRIVILLIG DELTAKELSE OG MULIGHET FOR Å TREKKE SITT SAMTYKKE

Det er frivillig å delta i prosjektet. Dersom dere ønsker å delta, tar dere kontakt med Wenche Ann Similä (se kontaktinformasjon under) og undertegner samtykkeerklæringen på siste side. Dere kan når som helst og uten å oppgi noen grunn trekke samtykket. Dette vil ikke få konsekvenser for videre behandling av barnet ditt. Dersom du trekker ditt barn fra prosjektet, kan du kreve å få slettet innsamlede prøver og opplysninger, med mindre opplysningene allerede er inngått i analyser eller brukt i vitenskapelige publikasjoner. Dersom du senere ønsker å trekke deg eller har spørsmål til prosjektet, kan du kontakte sykepleier, M.Sc. Wenche Ann Similä, epost wenche.ann.similae@stolav.no, telefon 72 82 56 27 eller prosjektleder og barnelege Torstein Baade Rø, epost torstein.ro@ntnu.no, telefon 72 57 40 51.

HVA SKJER MED INFORMASJONEN OM DITT BARN?

Informasjonen som registreres om barnet skal kun brukes slik som beskrevet i hensikten med studien. Du har rett til innsyn i hvilke opplysninger som er registrert om barnet ditt og rett til å få korrigert eventuelle feil i de opplysningene som er registrert.

Alle opplysningene vil bli behandlet uten navn og fødselsnummer eller andre direkte gjenkjennende opplysninger. En kode knytter barnet til sine opplysninger gjennom en navneliste.

Prosjektleder har ansvar for den daglige driften av forskningsprosjektet og at opplysninger om barnet blir behandlet på en sikker måte. Informasjon om barnet vil bli anonymisert eller slettet senest fem år etter prosjektslutt.

GODKJENNING

Prosjektet er godkjent av Regional komite for medisinsk og helsefaglig forskningsetikk, REK 2017/xx.

SAMTYKKE TIL DELTAKELSE I PROSJEKTET

JEG ER VILLIG TIL Å LA MITT BARN DELTA I PROSJEKTET

Som foresatte til_ (Fullt navn) samtykker vi til at hun/han kan delta i prosjektet

Sted og dato

Foresattes signatur

Foresattes navn med trykte bokstaver

Sted og dato

Foresattes signatur

Foresattes navn med trykte bokstaver

Jeg bekrefter å ha gitt informasjon om prosjektet

Sted og dato

Signatur

Rolle i prosjektet

INFORMASJONSSKRIV TIL BARN/UNGDOM 12-16 ÅR

HELSE HOS UNGDOM MED LANGVARIG UTMATTELSE

BAKGRUNN OG HENSIKT

Du får dette informasjonsskrivet fordi vi ønsker at du deltar i et forskningsprosjekt om ungdom med langvarig utmattelse/slitenhet. Vi vet for lite om hvordan det går med ungdom som er langvarig utmattet, og det er årsaken til at vi spør deg om å bli med i dette forskningsprosjektet. Vi håper at informasjonen du og andre ungdommer gir i prosjektet vil føre til at vi kan gi ungdom med langvarig utmattelse et bedre tilbud.

HVA INNEBÆRER STUDIEN?

Deltakelse i prosjektet medfører at du må svare på et spørreskjema om hvordan du har det og din helse. Dette vil ta 45-60 minutter. Videre ønsker vi også å intervjue deg på telefon eller på nærmere avtalt sted, dette vil ta ytterligere inntil 60 minutter. I tillegg henter vi opplysninger om utredningen hos oss fra din pasientjournal.

MULIGE FORDELER OG ULEMPER

Fordelen med deltakelse er at vi kan forbedre tilbudet til ungdom med langvarig utmattelse. Dette vil komme fremtidige pasienter til gode. Du vil få tilbud om en samtale med sykepleier etter intervjuet hvor du kan få råd vedrørende videre oppfølging, hvis du ønsker/trenger det. Ulempene ved å delta er at det vil ta tid for deg å svare på spørreskjema og delta i intervjuet. Vi vil legge til rette for at intervjuet kan skje på en måte og et tidspunkt som passer deg slik at du ikke behøver å ta fri fra skolen.

HVA SKJER MED PRØVENE OG INFORMASJONEN OM DEG?

Informasjonen blir registrert og lagret på en sikker måte, og bare helsepersonell som deltar i prosjektet vil ha tilgang til informasjonen. Informasjonen er avidentifisert, det betyr at opplysningene om deg blir behandlet uten navn og fødselsnummer. Informasjonen om deg blir slettet fem år etter prosjektslutt.

DELTAKELSE

Det er frivillig å delta i prosjektet. Du kan når som helst og uten å oppgi noen grunn trekke deg fra prosjektet. Dette vil ikke få konsekvenser for din videre behandling. Dersom du har spørsmål eller ønsker å trekke deg kan du kontakte Wenche Ann Similä, epost wenche.ann.similae@stolav.no, telefon 72 82 56 27.

NTNU
Kunnskap for en bedre verden

Vil du delta i forskningsprosjektet

«Læreres, rådgiveres og helsesykepleieres erfaringer fra arbeid med å legge til rette for en god skolehverdag for elever med kronisk utmattelsessyndrom»

Dette er et spørsmål til deg om å delta i et forskningsprosjekt hvor formålet er å undersøke skolepersonales erfaringer fra arbeid med å legge til rette for en god skolehverdag for ungdom med kronisk utmattelsessyndrom. I dette skrivet gir vi deg informasjon om målene for prosjektet og hva deltakelse vil innebære for deg.

Formål
Formålet med studien er å undersøke læreres, rådgiveres og helsesykepleieres erfaringer fra sitt arbeid med å legge til rette for en god skolehverdag for elever med kronisk utmattelsessyndrom i ungdomsskole og videregående skole. Studien er en del av et ph.d.-prosjekt som undersøker livskvalitet og skolefunksjon for ungdom med kronisk utmattelsessyndrom.

Hvem er ansvarlig for forskningsprosjektet?
Studien utføres av Norges Teknologiske Naturvitenskapelige Universitet (NTNU). Ph.d. stipendiat Wenche Ann Similä har ansvar for gjennomføring av datainnsamling. Torstein Baade Rø (Overlege og førsteamanuensis) er hovedveileder for studien og Torunn Hatlen Nøst (Sykepleier og Førsteamanuensis) er biveileder. Alle er ansatte ved NTNU.

Hvorfor får du spørsmål om å delta?
Du får spørsmål om å delta fordi du jobber i ungdomsskole eller videregående skole og har erfaring med ungdom med kronisk utmattelsessyndrom.

Vi har fått kontaktinformasjonen din fra:

……..
Det er innhentet godkjenning til å gjennomføre prosjektet fra Trøndelag fylkeskommune.

Hva innebærer det for deg å delta?
Deltagelse i studien innebærer å møte til et fokusgruppeintervju, og dele dine erfaringer fra arbeid med ungdom med kronisk utmattelsessyndrom i skolen. Du vil møte 5-7 andre lærere, rådgivere og helsesykepleiere fra andre skoler med lignende erfaring. Det vil gjøres lydopptak av samtalen i fokusgruppen. For å delta i studien skriver du under på samtykkeerklæringen på side 3 og returnerer den i vedlagte svarkonvolutt. Hvis du samtykker til å delta, vil vi ta kontakt med deg for å avtale tidspunkt for fokusgruppeintervjuet. En påminnelse om studien vil bli sendt deg på e-post hvis vi ikke hører noe fra deg innen 14 dager.

Det er frivillig å delta
Det er frivillig å delta i prosjektet. Hvis du velger å delta, kan du når som helst trekke samtykket tilbake uten å oppgi noen grunn. Alle dine personopplysninger vil da bli slettet. Det vil ikke ha noen negative konsekvenser for deg hvis du ikke vil delta eller senere velger å trekke deg.

NTNU
Kunnskap for en bedre verden

Ditt personvern – hvordan vi oppbevarer og bruker dine opplysninger
Vi vil bare bruke opplysningene om deg til formålene vi har fortalt om i dette skrivet. Vi behandler opplysningene konfidensielt og i samsvar med personvernregelverket.

- Alle opplysningene vil bli behandlet uten navn, fødselsnummer, adresse, skoletilhørighet eller andre direkte gjenkjennende opplysninger.
- En kode knytter deg til dine opplysninger gjennom en navneliste som lagres atskilt fra øvrige data. Utskrift av lydopptak vil ikke inneholde navn på deltagere eller skoler.
- Wenche Ann Similä, Torstein Baade Rø og Torunn Hatlen Nøst, alle NTNU, vil ha tilgang til innsamlede data.
- Det vil ikke være mulig å identifisere deg i resultatene av studien når disse publiseres.

Hva skjer med opplysningene dine når vi avslutter forskningsprosjektet?
Prosjektet skal etter planen avsluttes innen desember 2021. Bearbeidelse og publisering av data vil kunne skje innen to år etter prosjektets slutt. Datamaterialet anonymiseres ved utskriving av lydopptak. Lydopptak oppbevares på kryptert datafil og slettes etter transkribering, senest 31.12.2021.

Dine rettigheter
Så lenge du kan identifiseres i datamaterialet, har du rett til:

- innsyn i hvilke personopplysninger som er registrert om deg.
- å få rettet personopplysninger om deg,
- å få slettet personopplysninger om deg,
- å sende klage til Datatilsynet om behandlingen av dine personopplysninger.

Hva gir oss rett til å behandle personopplysninger om deg?
Vi behandler opplysninger om deg basert på ditt samtykke.

På oppdrag fra NTNU har NSD (420197) – Norsk senter for forskningsdata AS vurdert at behandlingen av personopplysninger i dette prosjektet er i samsvar med personvernregelverket.

Hvor kan jeg finne ut mer?
Hvis du har spørsmål til studien, eller ønsker å benytte deg av dine rettigheter, ta kontakt med:

- NTNU ved Wenche Ann Similä på epost *wenche.a.simila@ntnu.no* eller telefon: 99401830 eller Torstein Baade Rø på epost *torstein.ro@ntnu.no*.
- Vårt personvernombud: Thomas Helgesen, NTNU *thomas.helgesen@ntnu.no*

Hvis du har spørsmål knyttet til NSD sin vurdering av prosjektet, kan du ta kontakt med:

- NSD – Norsk senter for forskningsdata AS på epost (personverntjenester@nsd.no) eller på telefon: 55 58 21 17.

Med vennlig hilsen

Torstein Baade Rø
(Forsker/veileder)

Wenche Ann Similä
(Ph.d.-stipendiat)

NTNU
Kunnskap for en bedre verden

Samtykkeerklæring

Jeg har mottatt og forstått informasjon om prosjektet **«Læreres, rådgiveres og helsesykepleieres erfaringer fra arbeid med å legge til rette for en god skolehverdag for elever elever kronisk utmattelsessyndrom – en kvalitativ studie», og** har fått anledning til å stille spørsmål.
Jeg samtykker til:

- ☐ å delta i fokusgruppeintervju

Jeg samtykker til at mine opplysninger behandles frem til prosjektet er avsluttet

---

(Signert av prosjektdeltaker, dato)

Appendix 3

Data collection tools

List of Questionnaires, additional questions and data collection tool for data from medical records for Study 1, and interviewguides for Studies 2 and 3

List of Questionnaires

- *Pediatric Quality of Life Inventory ™ Generic Core Scale, version 4.0 (PedsQL4.0)* (255) https://www.pedsql.org/about_pedsql.html

- *The Pediatric Quality of Life Inventory™ Multidimensional Fatigue Scale (PedsQL-MFS)* (256) https://www.pedsql.org/about_pedsql.html

- *The Short Mood and Feelings Questionnaire (SMFQ)*(257).

- *De Paul Pediatric Health Questionnaire (Norwegian version)*(13, 24, 258).

(We also used EQ-5D-Y and EQ-5D-5L https://euroqol.org/
The results from these questionnaires are not included in this Thesis)

Spørsmål om oppfølging fra skole og helsepersonell

Har du hatt:	**JA**	**NEI**
Egen ukeplan		
Egen prøveplan		
Hjemmeundervisning		
Tilrettelagt timeplan		
Oppfølging fra lærer		
Oppfølging fra rådgiver i skolen		
Oppfølging fra PPT		
Oppfølging fra sosionom		
Oppfølging fra ernæringsfysiolog		
Oppfølging fra psykiatrisk sykepleier		
Oppfølging fra sykepleier		
Oppfølging fra helsesøster		
Oppfølging fra psykomotorisk fysioterapeut		
Oppfølging fra fysioterapeut		
Oppfølging fra ergoterapeut		
Oppfølging fra BUP		
Oppfølging fra fastlege		
Ansvarsgruppe		
Vært på rehabiliteringsopphold		
Vært på lærings og mestringskurs		
Mottatt hjelpemidler		
Har du forsinket skolegang		
Deltar du i fritidsaktiviteter		

IDnr: Dato:	«Helse og helserelatert livskvalitet hos ungdom med kronisk utmattelse»

Data fra Journal

Kjønn (gutt/jente/annet)	Gutt	Jente	Ubestemt		
Alder (hele år) v Diagnose					
Varighet av utmattelse ved utredningstidspunkt (måneder)					
Debut av utmattelse : (kan sette flere kryss)	Akutt	Gradvis	Post Inf.	Vaksine	Annet
Tid etter utredning (antall måneder)	Når diagnosen er satt. (mnd og år)		Dato i dag (mnd og år)		
Historikk fra årstall	Fra fødsel		Tilflytter (fra år)		

Antall polikliniske konsultasjoner i spesialisthelsetjenesten:	Før antatt sykdomsdebut relatert til kronisk utmattelse (Fødsel–2år før Dx)	**2 år før diagnose og fram til diagnosetidspunkt** (2år – Dx)	Etter diagnose
Lege			
Poliklinisk:			
Innleggelser + Liggedøgn			
BUP: * Kontakt med BUP (ja/nei) * Antall behandlingsepisoder			

* Diagnoser			

Diagnose ved innleggelse eller poliklinikk	Innleggelse	Poliklinisk

Kriterier for diagnose	Jason/KRUS
	Canada
	Fukuda
	Andre
	Ikke oppgitt

Felleskonsultasjon BUK-BUP (ja/nei)	JA	NEI	IKKE OPPGITT

Helsepersonell involvert i utredning:	Hak av i feltet nedenfor	Evt kommentar,
Lege		
BUP		
Sykepleier		
Ernæringsfysiolog,		
Fysioterapeut		
Ergoterapeut		
Pedagog		
Sosionom		
Andre		

Avholdt ansvarsgruppemøte (ja/nei)	JA	NEI	IKKE OPPGITT

(Se etter om det er tidligere sykdommer som går igjen hos flere og lage statistikk av dette i resultater)	

Andre aktuelle sykdommer ved eller etter diagnosetidspunkt			
Allergier			
skolefravær	<50%		
	>50%		
alvorlighet av utmattelse/CFS/ME ihht. Helsedirektoratets veileder (5).	Grad 1		
	Grad 2		
	Grad 3		
	Grad 4		
	Ikke spesifisert		
Bruk av medisiner ved diagnosetidspunkt (Nei/Ja, evt hvilke)	Ja	Nei	Spesifiser om ja:
Andre familiemedlemmer med CFS/ME?	Ja	Nei	Spesifiser hvem:
Gjennomgått EBV IgG IgM	JA	NEI	IKKE OPPGITT
Gjennomgått CMV IgG og IgM	JA	NEI	IKKE OPPGITT
Annet			

Intervjuguide for intervju med ungdom med CFS/ME
Del 1: Utmattelse

1. Opplever du for tiden noen problemer med utmattelse eller tretthet?
Hvis ja; beskriv symptomene
Hvis nei; hvor lenge er det siden du sist hadde disse plagene?

2. Hva kaller du det du har/har hatt?
Her er vi ute etter navn i form av diagnose eller andre betegnelser på tilstand og symptomer (CFS/ME, bare ME, kronisk utmattelse, slitenhet, utmattelse eller annet). Bruk personens betegnelse på symptomer senere i intervjuet

3. Har du hatt perioder hvor du har vært bedre eller dårligere enn du er nå?
Beskriv. Har forløpet vært svingende, gradvis verre, gradvis bedre, stabilt?
Illustrer gjerne ved en tidslinje. (evt sette inn en type tabell her også for gode og dårlige perioder?)

4. Hvordan har du opplevd det å være sliten/syk/utmattet/dårlig.
Beskriv hvordan det oppleves og føles.
Få tak i opplevelsen, i personens egne ord

5. Hvordan opplever du andres holdninger når du er/har vært sliten/syk/utmattet/dårlig
holdninger i familie, venner, skole, helsepersonell

6. Opplever du at andre tror på deg når du forteller hvordan du har/har hatt det?

	Ikke i det hele tatt	**Litt/av og til**	**Det meste av tiden**	**Alltid /nesten alltid**
Familien				
Venner				
Lærere/skole				
Helsepersonell				

7. Opplever du at andre forstår hva du strever/har strevd med?

	Ikke i det hele tatt	**Litt/i liten grad**	**Noe/til en viss grad**	**Veldig godt/i stor grad**
Familien				
Venner				
Lærere/skole				
Helsepersonell				

8. Har du noen å snakke med om vanskelige ting? Hvem? Hjelper det?

9. Er det andre i familien din som har vært eller er plaget med kronisk utmattelse?
- hvem?

- hvilken sykdom? (kronisk utmattelse kan komme av ulike sykdommer)

10. Har du fått andre sykdommer eller plager etter utredningen ved St. Olavs Hospital?

11. Hvor mye skolefravær (jobbfravær) hadde du i de 4 ukene før utredningen ved St Olavs Hospital (timer/dager)?

12. Hvor mye skolefravær (jobbfravær) har du hatt siste 4 uker (timer/dager)?

Skolefravær	4 uker før utredning	Siste 4 uker før intervju/studie
Timer per uke		
Dager per uke		

Del II: Helsehjelp og annen støtte

13. Får du eller har du fått hjelp fra noen for utmattelsen/trettheten?

- fra hvem?
- jevnlig?
- med hva?
- hjalp det?
- hvis ja, på hvilken måte hjalp det? varighet av effekt?
- hadde du en ansvarsgruppe i kommunen som møtte/møter deg eller dine foreldre?

Intervjuer krysser av på vegne av informanten:

MOTATT HELSEHJELP	Fra hvem (kryss av)	Hyppighet	Effekt	Varighet effekt
-Helsesøster				
-Fastlege				
-Ergoterapeut				
-Fysioterapeut				
-Psykolog				
-Sosionom				

-Annen helseperson i kommunen				
-PPT -Lærer/skole				
-Spesped i skolen				
-Rådgiver i skolen				
-Ansvarsgruppe				
I SYKEHUS	*	*	*	*
-lege				
-psykiater/-olog				
-fysioterapeut				
-ergoterapeut				
-sykepleier				
-ernærings-fysiolog				
-lærer				
-sosionom				
- Andre				

- hvor fornøyd er du med det tilbudet du har/har hatt på en skala fra 0-6 hvor 0=svært misfornøyd og 6 = svært fornøyd?

Skala fornøydhet:
(Svært misfornøyd) 0 1 2 3 4 5 6 (svært fornøyd)

- er det noe du savner/savnet i det tilbudet du har/ har hatt?

14. (Har du hatt kontakt med fastlegen siden du fikk utmattelsen?

Hvis ja; ca hvor ofte?)

Dette spørsmålet vil bli overflødig dersom vi bruker tabellen ovenfor

15. Har du søkt om hjelpemidler eller økonomisk støtte i forbindelse med sykdommen?

Hvilke hjelpemidler og/eller økonomisk støtte mottar eller har du mottatt?

Har du hatt ekstra egenutgifter på grunn av sykdommen?

	Fra hvem (kryss av)	Antall hjelpemidler og hvilke/hvilken økonom. støtte	Hyppighet	Har det hatt nytte/effekt
Mottatt hjelpemidler				
Mottatt økonomisk støtte				
Egne utgifter til nødvendig utstyr				

16. Har du / har du hatt hjelp fra noen i familien?

- hvem?
- til hva?
- har det hjulpet deg?
- hvis ja, på hvilken måte?

Familiær støtte	Hvem (kryss av)	Til hva	Hyppighet	Nytte/effekt
Mor				
Far				
Søsken				
Besteforeldre				

Tanter og onkler				
Andre				

17. Tror du at du selv kan påvirke slik at du blir bedre eller føler deg bedre?

– for eksempel med hva du gjør eller ikke gjør eller hvordan du tenker?

Ikke i det hele tatt	**Litt/i liten grad**	**Noe/til en viss grad**	**I stor grad**

18. Hva gjør du eller tenker du at du kan gjøre selv for å bli bedre eller føle deg bedre?

- hva?
- har noe hjulpet? Hva?

19. Har du gjort deg tanker om hva eller hvem som kunne ha hjulpet deg/kan hjelpe deg med å få det bedre?

20. Har du forsøkt noen form for psykologisk terapi eller annen terapiform?

- helsepersonell (lege/psykolog/psykiater/annen behandler)
- hvor hyppig?
- hjalp det? – på hvilken måte? (informantens opplevelse)

Terapi hos	Terapiform	Hyppighet	Nytte/effekt	Varighet effekt

21. Har du forsøkt alternativ medisin eller behandling?

- hvilken type? - hjalp det deg?

Alternativ behandling	Behandlingsform	Hyppighet	Nytte/effekt	Varighet effekt

22. Oppfølging og tilrettelegging i skolen:

- hadde du faste tider du møtte på skolen?
- hadde du tilrettelegging av undervisning?
- hjemmeundervisning, -undervisning i grupper?
- egen ukeplan for undervisning, -egen prøveplan?
- hjemmeoppgaver og muntlige prøver i stedet for skriftlige prøver med resten av klassen?
- fikk du tid til å prioritere sosiale aktiviteter på skolen?
- hadde du og/eller dine foreldre jevnlige møter med kontaktlærer og skole?
- ble det søkt om fritak i fag, fritak fra karakter i fag?
- hadde du kontakt med PPTjenesten ved skolen?

Tilrettelegging i skole med	Fra når	Hvor lenge	Regelmessighet/ Innhold	Manglende oppfølging
Faste tider				
Antall fag				
Tilrettelagt undervisning i skolen				
Hjemmeundervisning				
Egen ukeplan/prøveplan				
Hjemmeoppgaver/muntlige prøver				
Sosial plan i skolen				
Møter med skolen				
PPT søkt fritak (fra karakter i fag, fra hele faget)				
Annet				

23. Har du hatt kontakt med andre som plages med kronisk utmattelse?
(facebook, sosiale medier, pasientforening)

24. Har du deltatt eller deltar du i et annet forskningsprosjekt?

25. Har du deltatt på lærings- og mestringskurs?

26. Hvordan ser du for deg at tiden framover blir? (funksjon i skole, jobb, fritid)

27. Har du noe annet du ønsker å fortelle meg om *……(personens egne ord, f.eks utmattelsen el ME)* **eller oppfølgingen du har hatt etter at du ble syk?**

Spørsmål kun til foreldre:

Har sykdomsperioden hatt økonomiske konsekvenser for deg eller familien din?

Spørsmål til ungdom med CFS/ME - etter Covid-19 nedstengning

I forbindelse med digital undervisning på grunn av Covid-19 ønsker vi å stille oppfølgingsspørsmål til de som deltok på intervju. Dette gjøres ved å ta kontakt per telefon. Vi ønsker å stille spørsmål om hvordan ungdommene vi intervjuet har opplevd tilrettelagt undervisning i Covid-19 perioden og om det har hatt betydning for kontakten med skole. Data vil brukes som supplement til den kvalitative delen av studien.

1) Hvordan har du opplevd (tilrettelagt) undervisning i Covid-19 perioden?
2) Har det hatt betydning for kontakten med skole?

Fokusgruppe med lærere, rådgivere og helsesykepleiere i ungdomsskole og videregående skole om erfaringer fra arbeid med å legge til rette for en god skolehverdag for møte med elever med kronisk utmattelsessyndrom

Intervjuguide fokusgruppe

Innledning til samtale ved moderator:

Forskning viser at kontakten med skole kan ha en positiv innvirkning på livskvaliteten til elever med kronisk utmattelsessyndrom. Disse elvene lever med mange utfordringer og faller ofte delvis eller helt ut av det inkluderende fellesskapet i skolen som skal fremme helse, trivsel og læring.

Det vi ønsker å snakke med dere om i dag er:

-hva dere har av erfaringer fra arbeid med elever med kronisk utmattelsessyndrom i forhold til skolehverdagen.

Jeg har forberedt noen tema og spørsmål jeg vil stille. Det er flott om dere vil supplere med det dere tenker er viktig, og kom gjerne med innspill til hverandre. Det vil bli tatt lydopptak av samtalen og det er viktig at vi snakker en og en av gangen for å gjøre det enklere å høre på det etterpå.

- Det er viktig at vi ikke nevner navn på enkeltelever under samtalen.
- Rekk gjerne opp hånda om dere er flere som vil si noe samtidig så styrer vi det i rekkefølge så godt vi får til.

Det første jeg vil snakke med dere om er:

1) **Hva opplever dere at ungdom med kronisk utmattelsessyndrom har utfordringer med i forhold til skolehverdagen?**

2) **Hva gjør dere som lærere, rådgivere og helsesykepleiere i forhold til disse utfordringene?**

Stikkord og oppfølgingsspørsmål ved behov:

- **Tiltak –**
- **Kommunikasjon med eleven –**
  (Hvordan opprettholder dere en jevnlig kontakt med elever over måneder og år når de ikke er til stede på skolen?)
- **Skoletilknytning –**
  (*Hvilke tiltak gjøres for at elevene skal være tilknyttet skolen når de ikke er til stede over lengre tid og hvilken effekt har det?)*
- **Forventninger til Elev/Familie –**
  (Hva forventes det at elev og familie skal bidra med i forhold til det å være tilknyttet skolen når eleven ikke kan være til stede?)

- **Elevmedvirkning fra medelever –**
  (*Hvordan kan elevmedvirkning fra medelever bidra til at elever med kronisk utmattelsessyndrom får økt tilknytning til skolen?)*

- **Kommunikasjon mellom skolepersonale –**
  (Hvordan kommuniseres disse elevenes utfordringer mellom skolepersonale som den enkelte eleven har kontakt med? Hva kan hindre kommunikasjon mellom skolepersonale?)

- **Taushetsplikt**
  (På hvilken måte kan taushetsplikten overfor eleven hindre kommunikasjon?)

- **Ensomhet**
  (Har dere gjort dere tanker om at elevene opplever ensomhet som et stort problem?)

Da går vi videre til neste tema som er **Utdanningsdirektoratets fagfornyelsesplan**. **Den ble innført nå i høst og ifølge fagfornyelsesplanen skal elever lære å forstå og være i stand til å påvirke faktorer som har betydning for mestring av eget liv.**

3) **Hvilken betydning kan dette få for ungdom med kronisk utmattelse?**

Stikkord og oppfølgingsspørsmål ved behov:

- **Kunnskapsgrunnlag –**
  (I forhold til det tverrfaglige faget livsmestring, hvordan kan dere få kunnskap om den enkelte elevens erfaringer med å møte utfordringer?)

- **Elevmedvirkning –**
  (*Hvordan kan elevmedvirkning bidra til økt kunnskap?)*

- **Kommunikasjon mellom faglærere –**
  (*Hvordan kan elevenes erfaringer med å møte utfordringer kommuniseres mellom faglærere jamfør at livsmestring skal være et tverrfaglig tema?)*

- **Vurderingsgrunnlag –**
  (*Hvor mye av vurderingsgrunnlaget baseres på det tverrfaglige temaet livsmestring i de ulike fagene?)*

- **Påkobling av barnevern-**

Det siste temaet er,

4) **Hvilken betydning har Covid -19 hatt i forhold til ungdom med kronisk utmattelsessyndrom og deres skolegang?**

Stikkord og oppfølgingsspørsmål ved behov:

- **Covid-19 påvirkning –**
  (*Hvordan har Covid-19 påvirket kontakten med elever med kronisk utmattelsessyndrom?)*

- **Digitale plattformer og elevkontakt –**
  (*Hvordan kan digitale plattformer bidra til en bedre kontakt med elever med kronisk utmattelsessyndrom?)*

- **Digitale plattformer og skoletilknytning –**
  (*Hvordan kan digitale plattformer bidra til økt tilknytning til skolemiljøet?)*

- **Digitalt gruppearbeid –**
  (*Hvordan kan digitalt gruppearbeid med andre medelever gjennomføres for elever med kronisk utmattelsessyndrom?)*

- **Fagpersoner i digitale elevgrupper –**
  (*Hvor mye bør lærer, rådgiver eller helsesykepleier delta i en digital elevgruppe hvor elever med kronisk utmattelsessyndrom er tilstede?)*

- **Andre tiltak fra skolen –**
  (Hvilke andre tiltak gjøres eller kan gjøres i forhold til skolehverdagen for disse ungdommene?)

Intervju med lærere, rådgivere og helsesykepleiere i ungdomsskole og videregående skole om erfaringer fra arbeid med å legge til rette for en god skolehverdag for møte med elever med kronisk utmattelsessyndrom

Intervjuguide individuelle intervju

Det jeg ønsker å snakke med deg om i dag er hvilke erfaringer du har fra arbeid med elever med kronisk utmattelsessyndrom i forhold til skolehverdagen. Jeg har forberedt noen tema og spørsmål jeg vil stille. Det er flott om du vil supplere med det du tenker er viktig. Det vil bli tatt lydopptak av samtalen. Det er viktig at du ikke nevner navn på enkeltelever under samtalen.

Det første jeg vil snakke med deg om er:

1) **Kan du først fortelle litt om i hvilke forbindelser du har møtt elever med kronisk utmattelsessyndrom?**

2) **Ofte så fortelles det om at ungdom med kronisk utmattelsessyndrom har utfordringer med skole og utdanning. Hva tenker du om det?**

   - **Har du selv møtt på situasjoner hvor dette har vært utfordrende for deg?**
     (kom gjerne med eksempel)
     Hva var det som gjorde det utfordrende?
     (kom gjerne med eksempel)
     Har det vært situasjoner eller episoder som du tenker tilbake på som at her lyktes dere? Hva var det i så fall som gjorde at du tenker at da gikk det bra?
     (kom gjerne med eksempel)

3) **I de situasjonene du har hatt ansvar for elever med kronisk utmattelsessyndrom, kan du si noe om hvem det er du samarbeider med da- både internt på skolen og fra andre tjenester**?

   Er det noe du tenker på som spesielt utfordrende med slike samarbeid? Hvorfor det i så fall?
   (kom gjerne med eksempel)
   Har du noen tanker om hvordan du skulle ønske at et slikt samarbeid var?
   Når er det samarbeid fungerer godt tenker du? Hva er det som i så fall gjør at det fungerer?

Stikkord og oppfølgingsspørsmål om tiltak i skolen ved behov:

- Tiltak –

- Kommunikasjon med eleven –
  (Hvordan opprettholder du en jevnlig kontakt med elever over måneder og år når de ikke er til stede på skolen?)
- Skoletilknytning –
  (*Hvilke tiltak gjøres for at elevene skal være tilknyttet skolen når de ikke er til stede over lengre tid og hvilken effekt har det?)*
- Forventninger til Elev/Familie –
  (Hva forventes det at elev og familie skal bidra med i forhold til det å være tilknyttet skolen når eleven ikke kan være til stede?)
- Elevmedvirkning fra medelever –
  (*Hvordan kan elevmedvirkning fra medelever bidra til at elever med kronisk utmattelsessyndrom får økt tilknytning til skolen?)*
- Kommunikasjon mellom skolepersonale –
  (Hvordan kommuniseres disse elevenes utfordringer mellom skolepersonale som den enkelte eleven har kontakt med? Hva kan hindre kommunikasjon mellom skolepersonale?)
- Taushetsplikt
  (På hvilken måte kan taushetsplikten overfor eleven hindre kommunikasjon?)
- Ensomhet
  (Har du gjort deg tanker om at elevene opplever ensomhet som et stort problem?)
- Påkobling av barnevern-

Da går vi videre til neste tema som er **Utdanningsdirektoratets fagfornyelsesplan**. **Den ble innført nå i høst og ifølge fagfornyelsesplanen skal elever lære å forstå og være i stand til å påvirke faktorer som har betydning for mestring av eget liv.**

4) **Hvilken betydning kan dette få/har dette fått for ungdom med kronisk utmattelse?**

Stikkord og oppfølgingsspørsmål ved behov:

- Kunnskapsgrunnlag –
  (I forhold til det tverrfaglige faget livsmestring, hvordan kan du få kunnskap om den enkelte elevens erfaringer med å møte utfordringer?)

- Elevmedvirkning –
  (*Hvordan kan elevmedvirkning bidra til økt kunnskap?)*

- Kommunikasjon mellom faglærere –
  (*Hvordan kan elevenes erfaringer med å møte utfordringer kommuniseres mellom faglærere jamfør at livsmestring skal være et tverrfaglig tema?)*

- Vurderingsgrunnlag –
  (*Hvor mye av vurderingsgrunnlaget baseres på det tverrfaglige temaet livsmestring i de ulike fagene?)*

Det siste temaet er,

5) **Hvilken betydning har Covid -19 hatt i forhold til ungdom med kronisk utmattelsessyndrom og deres skolegang?**

Stikkord og oppfølgingsspørsmål ved behov:

- Covid-19 påvirkning –
  (*Hvordan har Covid-19 påvirket kontakten med elever med kronisk utmattelsessyndrom?)*

- Har du opplevd mer og/eller bedre kontakt med elver?

- Digitale plattformer og elevkontakt –
  (*Hvordan kan digitale plattformer bidra til en bedre kontakt med elever med kronisk utmattelsessyndrom?)*

- Digitale plattformer og skoletilknytning –
  (*Hvordan kan digitale plattformer bidra til økt tilknytning til skolemiljøet?)*

- Digitalt gruppearbeid –
  (*Hvordan kan digitalt gruppearbeid med andre medelever gjennomføres for elever med kronisk utmattelsessyndrom?)*

- Fagpersoner i digitale elevgrupper –
  (*Hvor mye bør lærer, rådgiver eller helsesykepleier delta i en digital elevgruppe hvor elever med kronisk utmattelsessyndrom er til stede?)*

- Andre tiltak fra skolen –
  (Hvilke andre tiltak gjøres eller kan gjøres i forhold til skolehverdagen for disse ungdommene?)

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