

Increased interaction and procedural flexibility favoured participation: Study across European cohorts of preterm born individuals

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Abstract

Objective: To understand participation and attrition phenomena variability in European cohorts of individuals born preterm through in-depth exploration of the interplay of situational elements involved.

Methods: Multi-situated qualitative design, using focus groups, semi-structured interviews and collaborative visual methodology with a purposive sample of adults born preterm, parents and professionals ($n = 124$) from eight cohorts in seven European countries.

Results: Most cohort participants were motivated by altruism/solidarity and gratitude/sense of duty to reciprocate (only absent in adults aged 19–21), followed by expectation of direct benefit to one's health and knowledge amongst participating adults. Common deterrents were perceived failure in reciprocity as in insufficient/inadequate interaction and information sharing, and postal questionnaires. Combining multipurpose, flexible strategies for contact and assessment, reminders, face-to-face and shorter periodicity and not simply adding retention strategies or financial incentives favoured participation. Professionals main challenges entailed resources, funding and, European societal changes related to communication and geopolitical environment.

Conclusion: Retention would benefit from tailoring inclusive strategies throughout the cohorts' life cycle and consistent promotion of reciprocal altruistic research goals. Investing in regular interaction, flexibility in procedures, participant involvement and return of results can help mitigate attrition as well as considering mothers as main facilitators to participating children and impaired adults.

Keywords: European cohorts, Premature, Participation, Multi-situated qualitative study, Collaborative methods

Abbreviations

BE Belgium

DK Denmark

EPIBEL Extremely Preterm Infants in Belgium

EPICE/SHIPS Effective Perinatal Intensive Care in Europe/Screening to improve Health In very Preterm infants in Europe

IT Italy

NL Netherlands

NTNU LBW NTNU Low Birth Weight in a Lifetime Perspective (Norway)

POPS Project On Preterm and Small for gestational age infants (Netherlands)

PT Portugal

The Sibling study Adults Born Preterm Sibling (Finland)

1 Introduction

Population-based cohorts are commonly conducted epidemiological studies and powerful study design in public health research. The use of large population-based samples promotes representativeness and the ability to capture the impact of policies and programme interventions on health quality and equity [1–3].

Despite the unquestionable relevance of such studies [4], external and internal validity are threatened by the loss of participants, affecting representativeness and biasing the measured associations. Alongside an adequate cohort recruitment, the main faced challenge is maximizing retention. Studies may face considerable selection biases with losses of 20% participants [5–7]. Understanding the elements that influence participation and retention in various settings is therefore critical for the success of longitudinal research.

Most studies providing insight into maximizing participant retention lack geographical variability, relevant details and consistent descriptions of the adopted strategies. Inferential leaps or generalization to other populations and settings, and subsequent usefulness of similar strategies may thus differ [8–15]. Further primary research on participation is needed to expand the diversity of assessed populations, methods and settings [15]. Although behavioural decision-making is complex, fluid and situational and influenced by individuals' personal traits, lived experience and emotional response [16–18], a paucity of literature regarding points of view and motivations to participate in cohorts remains [19–25].

This study provides an in-depth understanding of the variability of participation and the interplay of motives, facilitators and disincentives for participation, considering lived experiences of participants and professionals involved in diverse European cohorts of individuals born very preterm (VPT:<32 weeks of gestation) and/or with very low birth weight (VLBW:<1500g).

2 Material and methods

2.1 Study design

We used an inclusive qualitative approach of flexible multi-situated methods comprising the concept of multi-sites and situated knowledge [26–28]. The study protocol, published in full detail elsewhere [(31)], provided a shared flexible framework which included a collaborative visual methodology (VideoStories) [(32;33)], focus groups (FGs) and individual semi-structured interviews. Partnering research teams selected and combined those most pertinent to their contexts and targeted participants, using a co-constructed guide of key-issues to approach with subtopics tailored by local teams to the specificities of their cohort studies (**Web material 4**). The epistemological principle of flexibility in implementation maximized inclusiveness and diversity of contributors to the study.

2.2 Participants and settings

Potential participants were contacted and enrolled in collaboration with cohorts management teams. To satisfy saturation criterion and socio-geographic heterogeneous balance, it was used a combined purposive sampling strategy. Multi-site sub-samples and their sizes varied therefore within the range pre-established by the shared protocol, totaling 124 key-actors from eight cohorts in Belgium, Denmark, Finland, Italy, Norway, Portugal and The Netherlands, comprising 37 professionals, 41 parents and 46 participants, including individuals who failed to respond to previous study waves (**Web material 1**).

2.3 Data collection

Data were collected between April 2018 and June 2020 by local research teams. Country/region's official language was used, except for the FGs of professionals in Finland and Norway which were conducted in English. All audio recorded data, including participant-generated VideoStories, were transcribed and translated to English (**Web material 1**).

2.4 Data analysis

Data were handed over to the coordination team in Portugal and submitted to a triangulation of phenomenological thematic analysis with discourse analysis. Both visual and verbal depictions were treated as narratives [(34;35;36)]. Data sub-sets were therefore sorted and categorized by hand using an emergent/inductive strategy. Thematic analysis was used to determine emerged patterns and representational axes across data sub-sets and relevant deviances unique to certain individuals or settings. Additional information to triangulate our findings was gathered via: internal survey to cohorts management teams on implemented strategies, procedures and participation (**Web material 2**); and from 370 participants via open-questions added to a simultaneous follow-up of the POPS cohort, the largest and longest under study. Final interpretative analysis emerged by the generic application of the mode of contents contingency.

3 Results

Results are organized in major cross-contextual and context-specific situational elements involved in participation and attrition phenomena identified at various levels and stages for the eight European cohorts under study. Quotes are displayed in **Web material 3**.

3.1 Overrepresentation of female gender

3.1.1 Cross-contextual

Reflecting the gender profile of participant engagement in these cohort studies, our findings are dominated by female gender's viewpoints (75%). The gender distribution of contributing adults participating in cohorts was: F= 25, M=20, non-binary gender=1, and of parents responding on behalf of their children was: F= 34, M=7.

3.2 Motivations

3.2.1 Cross-contextual

3.2.1.1 Altruism/solidarity

Altruism/solidarity emerged as the leading motivation to participate in these cohort studies. The positive feeling of contributing to improve medical knowledge and health care practices to the future benefit of others and society appeared as the main representational axis across narratives from parents ($n=40/41$) and adults born preterm ($n=34/46$) (Quotes 1, 2, 3).

3.2.1.2 Gratitude/sense of duty to reciprocate

Gratitude/sense of duty to reciprocate to healthcare professionals/scientific community or redirected to counterparts seconded altruism/solidarity in most narratives. Its concurrent expression prevailed amongst parents across child cohorts ($n=27/34$); became notoriously absent amongst the young adults in the context of EPIBEL cohort; and resurfaced amongst one fourth of their mothers and adults aged 30-39 (Quotes 4, 5, 6).

3.2.2 Context-specific

3.2.2.1 Expectation of direct benefit

The expectation of direct benefit to learn about prematurity, oneself, and others, replaced the relevance of gratitude/sense of duty to reciprocate amongst many adult cohort participants ($n=31/46$). It gained expression in more than half of younger adults narratives ($n=4/7$) and in more than 70% of those aged 30-39 (Quote 7).

As adults, face-to-face physical assessments were highly appraised ($n=27/37$). Two thirds of them added being particularly motivated by the expectation of direct benefit and positive health behaviour changes from health assessments (Quote 8).

As for the child cohorts, parents in 11 ($n=3$) and BE ($n=4$) pointed out the expectation of direct benefit for their children. They were not fully aware of the independence of EPICE/SHIPS studies from the clinical follow-up of their children while also voicing their frustration/distrust in the healthcare system (Quotes 9, 10). For all other parents ($n=27/34$), the expectation of any direct benefit was either absent or denied (Quote 11).

3.2.2.2 Being part of a researched group

The positive feeling of being part of a selected researched group was pointed out as important motivation by one EPIBEL participant and nearly half of the adults from POPS ($n=13/28$) (Quote 12).

Additional data collected from our added questionnaire to POPS follow-up allowed to contextualize the local representativeness of these findings for this whole cohort. Of 241 participants who contributed via open-ended responses on motivations: 82% framed their answers under altruism/solidarity; 32% also/or under the expectation of direct benefit; and 10% also/or under the positive feeling of being part of a selected researched group.

POPS is the largest and longest cohort. It stood out as the one promoting most varied interaction with participants over time. Nevertheless, participation significantly declined after participants reached adulthood (follow-up at 19years: 74%; at 28years: 34%; at 35years: 39%) in line with the decrease of frequency of interaction.

3.3 Motivational deterrents

3.3.1 Cross-contextual

3.3.1.1 Perceived irregular, insufficient/inadequate interaction and information sharing

A large majority of parents ($n=31/41$) and adults aged 30-39 ($n=27/37$), either when addressing major motivational deterrents or suggestions to improve participation, manifested a sense of dissatisfaction towards interaction and sharing of information along with a failed expectation of increased frequency, regularity and adequacy (Quotes 13, 14, 15).

In addition, half of the youngest adults that for the first-time provided consent on their own have specifically addressed the expectation of being kept well informed about the study and its findings to keep participating (Quote 16).

3.3.2 Context-specific

3.3.2.1 Perceived entanglement of clinical and research follow-ups

Most parents confessed not having retained information on either the research or its prospective trait at the enrolment of their newborn in EPICE/SHIPS due to distressful, overwhelming experience at the time ($n=29/34$). As recruitment occurred at the hospital unit before discharge, clinical and research follow-ups have been perceived by parents as intertwined, as part of the care package for their children. All parents, interviewed face-to-face ($n=28$), shared emotional accounts on trying to cope with mandatory extensive clinical appointments, therapies, and treatments along with the cohort solicitations. Targeted parents that failed to respond to follow-ups ($n=9$) added descriptions of being mother of twins or more children, severe child impairments, single parenthood and/or frustration/distrust in the healthcare system for not having responded adequately to their needs. When reasoning about the motives for their decision, non-response was explained by no surplus of energy or inattention due to their demanding lived experience as a mother. While some did not even remember not having responded, most declared that researchers should have insisted on obtaining their positive response ($n=7/9$) (Quotes 17, 18).

3.4 Situational elements related to studies strategic procedures

3.4.1 Cross-contextual

3.4.1.1 Combining multiple, flexible strategies

All cohort participants expressed appreciation for flexibility and alternatives offered by studies to contact and facilitate participation. It was a matter both of preference and of some not being at all approachable via particular methods. Combining email (75%) with phone (67%) and/or postal mail (39%) with reminders, emerged as the most favoured, while 63% recommended not to use postal mail alone. The use of social platforms, such as WhatsApp and Facebook, was explicitly repudiated by 17% of participating adults, including the youngest (Quotes 19, 20 and 21).

Most of adults born preterm (78%) further endorsed the combined use of multiple flexible/tailored methods to minimize non-response and attrition bias (Quotes 22, 23).

3.4.1.2 Increased frequency of interaction and face-to-face encounters

Both POPS after reached adulthood and EPICE/SHIPS cohorts were mainly assessed via questionnaires at a distance. The Finnish Sibling study and NTNU LBW Life heavily relied on face-to-face assessments combining

physical/medical examination with onsite administration of questionnaires. In EPIBEL, participants experienced both kinds of assessment twice.

More than half of participants aged 30–39 reported to expect further face-to-face assessments/interaction. That included all participants of The Sibling study who indicated having experienced long physical and emotionally demanding exams, including painful muscle biopsies for research purposes ($n=9/9$). In POPS, majority of participants also stated the failed expectation of shorter periodicity of interaction, including face-to-face events like the last one at the 19 years study wave ($n=17/28$) (Quote 24).

In all child cohorts, parents expressed their disappointment for the lack of initiatives to engage more with researchers and/or other participants. All parents who had participated in face-to-face assessments and gatherings reported these experiences as motivating ($n=34/41$) (Quote 25). Significantly, in EPICE/SHIPS-PT, all those who made improvement suggestions for study bonding also emphasized face-to-face interaction. According to them, it is the strategy *par excellence* to enable closeness and familiarity with the faces behind the study (Quote 26).

The relevance of these findings is reinforced when looking into the interplay of histories of interaction and participation trends across cohorts with comparable life cycle periods. NTNU LBW Life shows significantly higher retention of adult participants (76%) than POPS (39%), while resorting to apparently similar retention strategies over time. They differed greatly in periodicity and methods of interaction. As for EPICE/SHIPS, the PT cohort showing the highest retention (83%), stood out by its divergent strategies of extending the face-to-face assessment at 5 years to the whole cohort, yearly monitoring and birthday postcard sent to all children (**Web material 2**).

3.4.1.3 Postal questionnaires

Postal questionnaires acted as deterrents to participation. When looking closely to common study waves across child cohorts, we found postal questionnaires associated to poorer response in all four settings while even poorer in Belgium and Denmark, where no alternatives were offered. Some of the mothers in Denmark that had not responded to follow-ups via postal questionnaire, had participated in other research studies with their children ($n=3/7$). As they explained, the assessment method was the closing factor in the weighing process for their decision of non-response (Quote 27).

Participants recommended flexibility in administration and return of questionnaires, such as by phone, in electronic form/online or face-to-face, and that they are short and straightforward ($n=18/34$ parents; $n=23/46$ adults born preterm) (Quotes 28, 29, 30). Those favouring more personal approaches, to clarify doubts and to deepen contributions, further advised to opt for more interactive procedures (Quote 31).

Our additional data from the whole POPS cohort points once more to the representativeness of these findings. Out of those who responded on most dissatisfactory aspects: 71% framed their answers under insufficient/inadequate interaction and information sharing on the study and its findings ($n=66/93$); while 31% singled out insufficiency of face-to-face procedures and/or questionnaires inadequacy to include participants with varied attributes and/or to deepen contributions ($n=29/93$).

3.5 Situational challenges faced by professionals

3.5.1 Cross-contextual

3.5.1.1 Financial and human resources constraints

Losing study participants through failure to locate/contact or to respond due to burdensome/unsuitable follow-up procedures emerged as the major concern of professionals in all settings ($n=37/37$). Converging with cohort participants standpoints, professionals in all settings advocated the usefulness of: a) flexibility to reconcile study and participants agendas; b) enhancing proximity: alternative methods, locations and language mediators, monetary assistance/rewards; c) enhancing bonding: adequate information sharing, research team continuity (familiarity). However, all discussions raised situational challenges to implement such strategies due to financial and human resources constraints, though differing in severity across contexts.

Denmark, Finland and Norway have nationwide registries with personal identity numbers covering virtually all individuals residing in those countries and enabling data linkage to trace participants. Main faced challenge has been in selecting and implementing the most appropriate and inclusive strategies for interacting (Quotes 32, 33). Research teams in non-Nordic countries added to those challenges the extra effort and resources required just to keep track of participants (Quote 34).

In the EPIBEL and EPICE/SHIPS contexts, it was further exposed the dependency on short-term research funding which limited the possibilities to provide participants with prospective information. Consent to participate must be restricted to the protocol framed by the funded project and as such cannot anticipate long-term future interactions for which funding is not yet assured. It further constrained ensuring regular contact in-between study waves and team/staff continuity.

EPIBEL is exemplary on how the effect of these cumulative constraints severely impacted the ability to trace, interact and retain participants. Over its 20 years of existence, research teams were able to perform three follow-ups, the last two with a periodicity of 8–9 years while each time most resources were absorbed just to trace, re-invite and provide incentives to the particular event. Not only had the cohort follow-up been restricted from national to the Flanders region, it had also been limited to Dutch-speakers, though the country recognises three officially spoken languages and internal mobility and migration is common (Quote 35).

3.5.1.2 Societal changes related with communication systems

The fast rhythm of changes regarding communication in the last fifteen years has hampered the efficiency of available tracing systems. Though the impact seemed less evident for the adult cohorts in Nordic countries, EPICE/SHIPS-DK has faced similar challenges. Most influential elements reported were: a) the impact of progressive dismissal of home phone landlines and reliance on changeable mobile/electronic contacts detached from physical addresses; b) the increased informatization of databases and work processes with replacement of systems at times asynchronous and discordant; and c) legislations/regulations increasingly constraining access to personal data and record linkage (Quotes 36, 37, 38).

3.5.1.3 European Union geopolitical environment

Current geopolitical scenario related with population mobility, immigration and displacement has increased all cohorts vulnerability to existing logistical constraints. Though stressed in all discussions, these challenges were acuter in Italy and Belgium and in the context of child cohorts recruited in 2011–12. After being discharged and/or stopping clinical follow-up, particularly foreign immigrants and vulnerable families that moved frequently became increasingly difficult to trace and to be provided with context-sensitive alternatives to participate (Quote 39).

4 Discussion

Main shared motivations to participate in these cohorts were altruism/solidarity and gratitude/sense of duty to reciprocate. Major motivational deterrents were the perception of poor interaction and sharing of study findings. Combining multiple, flexible strategies of contact and assessment favoured participation while using postal questionnaires acted as deterrent. Main challenges faced by professionals were financial and human resources, and European societal changes related to communications, population mobility, immigration and displacement, hampering their ability to optimize tracing and interacting, especially with foreign-born and vulnerable families.

Our findings were dominated by females' viewpoints which reflected the gender profile of participant engagement in these cohort studies, particularly when involving parents of child participants. Though consideration of the child's willingness has a rights-based dimension, their parents' viewpoints and motivations are determinant to understand participation until consent and accountability is passed on [22;37;38;37;38]. Adult males were more likely to participate when research concerned themselves, and less when it targeted their children; and for those not fully autonomous/with impairments, mothers continued to be the ones more likely to facilitate participation. As traditionally found in other family and child development research, this gender imbalance in participant engagement suggests that the female parent viewpoint and lived experience on participation should be carefully considered in strategic management decisions [(39)].

Aside from altruism and gratitude, a majority of participating adults added the expectation of direct benefit by personally learning more about prematurity, themselves, and others. In POPS cohort, a significant number of individuals also pointed out being part of a selective researched group as important motivation. Insufficient interaction and information sharing emerged as main motivational deterrent in all settings. Thus, irrespectively of the underlying motivations to endure participating, the researcher stance has been perceived as failing in reciprocity.

Motivations of altruism/solidarity have been pointed out as underlying factors to enrol in and to endure participating in longitudinal studies [8,20,40,41]. Our findings are similar to previous observations in what appears to be the paradoxical concurrence of this motivation with others. Some authors interpreted it through combined concepts such as conditional altruism, weak altruism or perhaps less truthful to be more socially acceptable [(42;43)]. Child cohorts under study did not presuppose any kind of incentives. It is therefore reasonable to interpret their participation as an act of gift-giving, of generous transfer of socially valued objects without any guarantee of reciprocation. Amongst adults, though financial incentives for assessment completion have been used at times, the overwhelming majority did not refer to them as a suggestion to improve participation and all refuted their role in retention.

The complete absence of gratitude/sense of duty to reciprocate as a concurrent motivation occurred as a deviance particular to the cohort of adults aged 19–21 years. Verbal expression of connective gratitude tends to increase with age [(44;45)], which our findings reflected. Those more prone to feel gratitude seem to have more neural hallmarks of altruism and other brain areas associated with feelings of reward when contributing to the benefit of others [(46;47)]. As gratitude is associated with acknowledgement and the most significant form of acknowledgement is return [(54;55;56;57)] this finding suggests that researchers should focus in displaying reciprocal altruism during the cohort's

transition to adulthood to ensure a prospective balanced relationship. Its promotion may be especially useful for planning research with adolescents born preterm with special needs or social adjustment difficulties because gratitude relates to personal well-being and social relationships satisfaction [(48;49;50;41;52;53)].

In the context of our European child cohorts, phenomena of participation and attrition were linked to parents' lived experiences on having a preterm birth and on the support provided by the healthcare system. Parents' vulnerability and distressing experiences at the time of enrolment favoured receptivity to being researched while finding comfort in contributing to the benefit of others [(58;59;60)]. In the long-term however, the continued exposure to distressful experiences of parenthood when combined with perceived lack of healthcare support or effort-reward imbalance led some to increased carelessness or non-response to cohort solicitations. We also found that even parents describing healthy children and stable family support shared the perception of inadequate psychological healthcare provision to mothers of VPT children. These results suggest that aspects of prematurity such as psychological distress and general parental stress and coping, either have not yet received enough research attention [(62;63;64)] or findings are not being adequately translated into healthcare policies and practices in Europe.

All cohorts implemented multiple strategies specifically aiming to maximize retention but range and diversity of procedures differed. As shown in **Web material 2**, diversity and frequency of applied retention strategies did not allow to identify a straightforward trend on their effectiveness. To understand variability in participation required to look further into cohorts' study designs, how strategic procedures were modified over time, and what was the interplay of lived experiences.

We identified major situational facilitators and deterrents related to strategic procedures. However, combining multiple alternatives and flexible/tailored strategies aiming to reduce participant burden favoured participation; while merely adding more retention strategies or monetary incentives did not seem to result in higher retention, as shown in a review of cohort studies in general [9]. When it comes to cohorts of VPT/VLBW individuals, a recent systematic review ($n=39$) reported that retention strategies were rarely described in the publications, hampering the assessment of such strategies on retention, which highlights the need for further context-sensitive primary research like the present study [(65)].

All eight cohorts faced the impact and challenges of influx and mobility of population of the last decade within geopolitical European context, whereas Italy and Belgium stand out for the much higher increase of foreign vulnerable population density [(66;67;68;69)]. Professionals in all settings shared long reported concerns that people from minority and vulnerable groups are at higher risk of loss to follow-up in cohort studies [(70;71;72;73;74;75;76)].

4.1 Limitations

Access to individuals that had withdrawn consent to cohort participation was unattainable. Limited information could be retrieved from male informants.

5 Conclusion

This multi-situated collaborative study provided valuable insights to guide researchers in European cohorts of individuals born preterm to improve participation, which would benefit from the consistent promotion of reciprocal altruistic research throughout the cohort's life cycle. The ultimate benefit is however the potential transformative impact on participants. As well known, preterm birth is associated with various impairments and morbidities, increased anxiety, social rejection and reduced self-esteem. Results suggest that enhancing frequency and regularity of interaction and information sharing through flexible, inclusive strategies tailored in-context to participants might be most effective in maximising retention. Challenging geopolitical and socio-economic environments combined with inadequate funding restrict the possibilities to implement desired strategies to optimise response. We were able to find that, instead of adding retention strategies, opting for multipurpose more inclusive ones may help in calibrating cost-effectiveness.

5.1 Practical implications

Participation in European cohort studies of individuals born preterm may benefit from: planning protocols open to flexibility and revision to incorporate in-context tailored strategies that may offer greater inclusiveness throughout the cohorts' life cycle; regular interaction and information sharing, participant involvement, and consideration of mothers as main facilitators to participating children and impaired adults. Further effort should be made in identifying male and non-binary gender's research expectations.

Authors contributions

SCSM contribution to the conceptualization and implementation of the study, data collection, analysis and first draft of this manuscript. JD contributed to the implementation of the study, data collection, data analysis, draft of this manuscript, review and editing of this manuscript. RT contributed to the data analysis, draft, review and editing of this manuscript. GA, AB, AMB, GC, MC, KAIE, VH, MSI, EK, JL, SP, PP, IS and EV contributed to implementation and

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Data availability statement

Multi-site interview transcripts datasets generated for this study cannot be shared for legal, ethical and privacy restrictions. In accordance with multi-site ethical clearances and signed informed consent provided by participants which guarantees their anonymity and confidentiality, generated data for this study may only be accessed and handled within RECAP pPreterm research team and under the framework of internal governance of the Horizon 2020 project RECAP pPreterm funded by the European Union under grant agreement N° 733280.

Ethical approval

Approval by Ethics Committees, Data Protection Authorities and signed written informed consents by all responders in their spoken languages were obtained according to national rules. As required, ethical clearance was obtained from the Ethics Committee of Antwerp University Hospital in Belgium; the Danish Ethical Committee System and Danish Data Protection Agency in Denmark; the Ethics Committee of Hospital District of Helsinki and Uusimaa in Finland; the Ethics Committee of Paediatric Hospital Bambino Gesù in Italy; the Ethics Committee of the Institute of Public Health of the University of Porto and Portuguese Data Protection Authority in Portugal; and, the TNO Institutional Review Board in The Netherlands.

We confirm all patient/personal identifiers have been removed or disguised so the patient/person(s) described are not identifiable and cannot be identified through the details of the story.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.jclinepi.2021.12.027](https://doi.org/10.1016/j.jclinepi.2021.12.027).

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