DOUTORAMENTO EM SAÚDE PÚBLICA GLOBAL

Quality of life in parents of very preterm infants: insights from family integrated care

Mariana Filipa da Silva Amorim

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Abbreviations

ART	Assisted Reproductive Technologies
EDI	European Deprivation Index
ESRI	Environmental Systems Research Institute
EuroQoL	European Quality of Life Measure
FICare	Family Integrated Care
ISPUP	Instituto de Saúde Pública da Universidade do Porto
LOS	Length of stay
NICU	Neonatal Intensive Care Unit
NICUs	Neonatal Intensive Care Units
OECD	Organisation for Economic Co-operation and Development
PCO	Portuguese Classification of Occupations
QoL	Quality of Life
SF-36	Short-form Health Survey
UK	United Kingdom
USA	United States of America
WHO	World Health Organization

Abstract

Background

Preterm birth is a major public health priority. Preterm infants are at high risk of neonatal morbidity and its associated complications are one of the leading causes of global deaths among children under 5 years of age. A preterm childbirth and the ensuing infant's admission in a Neonatal Intensive Care Unit (NICU) affects family environment and quality of life (QoL) for many years. Most studies about the factors influencing the QoL of parents of preterm infants have excluded fathers and neglected the analysis of structural levels, offering a one-sided perspective that fails to assess dimensions related with shared governance for health, limiting the opportunities of co-producing family integrated healthcare systems. Furthermore, there is a paucity of sensitive instruments adapted to the singular experience of parenting a very preterm infant. This evidences the need to gain a better understanding about both parents' experiences, which can be achieved through mixed-methods research (**PAPER I**).

Exploring the QoL of parents of very preterm infants and its associated factors requires for a detailed analysis of their experiences, needs and sources of stress not only during the NICU hospitalisation, but also after discharge. Such in-depth perspectives will allow the identification of parents at risk for immediate and extended physical and emotional burden and the reduction of social inequalities and stress associated with caring for a very preterm infant.

Objectives

Focusing on family integrated healthcare, this thesis aims to broaden the knowledge on parental QoL following a premature birth to sustain the development of policy and practice in neonatology. This mixed methods study will explore sources of stress, needs and QoL of mothers and fathers of very preterm infants, serving as a foundation to address the following specific objectives:

1. To identify sources of stress in mothers and fathers of very preterm infants hospitalised in NICU, and their association with socio-demographic, obstetric and infant's characteristics.
2. To validate the NICU Family Needs Inventory for the Portuguese population, and to propose a Short Form.

3. To explore the needs of mothers and fathers of very preterm infants hospitalised in NICU according to their socioeconomic position, obstetric history and infant's characteristics by integrating quantitative and qualitative data.
4. To explore both mothers' and fathers' perspectives about their own QoL 4 months after a very preterm delivery, by integrating quantitative and qualitative data.

Methods

This is an observational and longitudinal mixed methods study. A multistage design was used, involving the collection of quantitative and qualitative data in two different time points: 1) during a NICU hospitalisation period, 15 to 22 days after childbirth, using individual quantitative questionnaires; 2) after discharge, approximately 4 months after childbirth, using qualitative semi-structured couples-based interviews and individual quantitative questionnaires.

Between the 1st of July 2013 and the 30th of June 2014, 120 mothers and 91 fathers of very preterm infants hospitalised in the 7 level III NICUs of the Northern Health Region of Portugal were systematically recruited (participation rate = 96.8%). Trained interviewers conducted face-to-face interviews, using structured questionnaires, to mothers and fathers separately but within the same timeframe. Data on demographic and socioeconomic characteristics as well as obstetric history was collected by self-report and clinical records of NICU hospitalisation were reviewed to retrieve data on pregnancy complications, mode of delivery, multiple pregnancy and neonatal characteristics of infants. Data on perceived social support, parental needs in NICU, and parental sources of stress were collected through self-administered questionnaires. Approximately 4 months after childbirth, between the 1st of November 2013 and the 30th of November 2014, self-administered questionnaires to be completed individually were sent by postal mail to 113 families who previously accepted to participate, and 67 mothers and 64 fathers completed and returned the questionnaires (participation rate = 59.3%). Data on infant's length of stay and diagnosed health problems were collected as well as symptoms of anxiety and depression among parents, parenting stress and perceived QoL. A subsample of 26 parental couples was jointly interviewed, between November 2013 and April 2014. Participants were purposively sampled to include parents of extremely and non-extremely low birth weight infants. A heterogeneity sampling was used for maximum variation of views and experiences, until reaching thematic saturation.

Descriptive and inferential statistics were used to analyse the quantitative data. In qualitative data, thematic content analysis was performed using a triangulation strategy.

Quotations with similar meanings were synthesized into categories, both inductively and deductively (according to the objectives of each paper), and then grouped into analytical themes.

Results

Parents classified the overall experience of infants' hospitalisation in a NICU as more stressful than the median for the remaining subscales. "Change in parental role" was classified as the most stressful source by mothers (Median (P25–P75): 4.1 (3.2–4.7)) and fathers (Median (P25–P75): 3.2 (2.4–4.0)). Mothers scored significantly higher in all subscales than fathers. For mothers, multiple pregnancy was associated with lower levels of stress regarding "change in parental role" ($\beta = -0.597$; 95% CI = −1.020 to −0.174) and "overall stress" ($\beta = -0.603$; 95% CI = −1.052 to −0.153). Being ≥30 years old was found to be negatively associated with fathers' overall stress and in all domains (**PAPER II**).

Exploratory factor analysis of the *Neonatal Intensive Care Units Family Needs Inventory* revealed two dimensions, one focused on the parents' needs and another on the infant's needs. To compose the Short Form Inventory, items with ceiling effect were eliminated and 22 items were submitted to confirmatory analysis, which supported the existence of two dimensions (CFI = 0.925). The Short Form showed a high degree of reliability (alpha ≥ 0.76). Less educated and older parents more frequently attributed a significantly higher importance to parent-centred needs, while parents of multiples revealed a tendency to value infant-centred needs (**PAPER III**).

Mothers valued information needs more than fathers (Median (IQR): 3.8 (3.6-3.9) vs. 3.7 (3.5-3.9)). First-time fathers, as well as older and less educated mothers reported more needs than younger and more educated participants. Despite gender differences, the assurance and proximity needs of parents apply across NICUs. Qualitative findings added the following needs: instrumental support from the government; regular emotional support from psychologists and social workers; enhancement of privacy in the neonatology ward to assure family-centred information and comfort; and availability of other parents and health professionals as complementary health mediators in the provision of detailed and coherent information (**PAPER IV**).

The perception of QoL was comparable to the scores observed in the Portuguese general population, and was not significantly different by gender. QoL scores increased slightly from the environment (Mean (SD): 72.1 (14.2)) to the psychological domains (Mean (SD): 78.7 (14.4)). Parenting stress, anxiety or depressive symptoms negatively influenced both maternal and paternal QoL. Lower socioeconomic position negatively influenced both parents' perceptions concerning the environment domain, and maternal physical and psychological QoL. Infant-related factors (extremely low birth weight, hospitalisation in the NICU for 2 months or more, or infant's health problems) were negatively associated with overall QoL among mothers and with the physical, psychological, social and environment domains among fathers. Qualitative findings indicated the activation of accommodation mechanisms by parents counterbalance constraining factors (surveillance, sleep disturbances, non-supportive healthcare policies, hygienization) with facilitating factors (social support, accessibility/quality of healthcare, opportunities for developing parental skills) of QoL. These processes were anchored on child-centredness and the construction of hierarchies of hope and expectations about infants' health and development (**PAPER V**).

Conclusion

This thesis provides evidence to sustain the development of policy and practice in family integrated neonatal services by analysing parental QoL following the birth of a premature infant. Taking a public health approach, it focuses on individual, familial, socioeconomic and political characteristics. Achievements suggested that factors related with sociodemographic characteristics and reproductive trajectory influenced parental stress and needs during the third week of infant's hospitalisation in a NICU, differently according to gender. Aspects related to infants and parental psychological characteristics were associated with parental QoL 4 months after the childbirth. Individual accommodation mechanisms and structural factors influenced parental QoL throughout the experience of parenting a very preterm infant.

This thesis adds to the conceptualization of family integrated healthcare five key areas: analysis of parental QoL as an outcome; gendered sensitive assessment of parental needs and sources of stress in NICUs; inclusion of both mothers' and fathers' reproductive trajectories and privacy within staff and unit as influencing factors; consideration for the role played by the community and extended family; and, introduction of the socioeconomic and political context in which infants and families live in (e.g. health governance, financing and resources; social assistance; labour; cultural and societal norms and values).

Findings also suggest the implementation of interventions focused on reducing parental stress and the disempowering effects of surveillance and hygienization on QoL to diminish disparities in family health. Moreover, integrated health services call for the guarantee of instrumental support from the government; regular emotional support from psychologists and social workers; enhancement of privacy in the neonatology ward to assure family-centred information and comfort; and availability of other parents and health professionals as complementary health mediators in the provision of detailed and coherent information. Finally,

this thesis recommends flexibility and sensitivity in research to develop instruments to assess sources of stress, parental needs, and parental QoL that take notice of gender, social support, socioeconomic position and reproductive trajectories of parents, as well as issues of privacy and regular emotional support in NICUs.

From a public health standpoint, this thesis raises issues that should be acknowledged on the co-production of intersectoral family-centred public policies, integrated healthcare services and focused-interventions to promote parental QoL. The comprehensive approach undertaken by this thesis renders its results applicable to various neonatal settings and contributes for rethinking governance in neonatology, by promoting the coordination of care both with and around the needs of infants, their families and communities.

Resumo

Introdução

A prematuridade é uma prioridade relevante em saúde pública, uma vez que as crianças nascidas prematuramente têm um risco elevado de morbilidade neonatal e as complicações daí resultantes representam uma das principais causas globais de morte antes dos 5 anos de idade. O nascimento de uma criança prematura e o consequente internamento numa Unidade de Cuidados Intensivos Neonatais (UCIN) afetam o agregado familiar e a qualidade de vida (QdV) dos seus elementos durante muitos anos. A maioria dos estudos acerca dos fatores que influenciam a QdV de pais de crianças prematuras exclui a perspetiva do pai e negligencia o nível de análise estrutural, proporcionando uma perspetiva limitada que não avalia dimensões relacionadas com a governação de saúde e restringe as oportunidades de coprodução de sistemas de cuidados de saúde integrados e centrados na família. Para além disso, verifica-se a ausência de instrumentos de avaliação sensíveis e adaptados à experiência de pais de crianças muito prematuras. Isso evidencia a necessidade de um melhor entendimento sobre a experiência parental, o qual pode ser alcançado através de investigação mista, ou seja, com recurso simultâneo a metodologias quantitativas e qualitativas (**ARTIGO I**).

Para compreender a QdV de pais de crianças muito prematuras e explorar os fatores que lhe estão associados, importa proceder a uma análise aprofundada das suas experiências, necessidades e fontes de *stress* não só durante o internamento na UCIN, mas também após a alta. Este conhecimento permitirá identificar os pais em risco, imediato ou futuro, de consequências físicas e emocionais, promovendo a redução das desigualdades sociais e do *stress* associados ao cuidar de uma criança muito prematura.

Objetivos

Com base numa abordagem de cuidados de saúde integrados e centrados na família, esta tese tem como objetivo ampliar o conhecimento acerca da QdV de mães e pais de crianças nascidas muito prematuramente para sustentar o desenvolvimento de políticas e práticas de saúde em neonatologia. Utilizando metodologias mistas, este estudo explorará as fontes de *stress*, necessidades e QdV de mães e pais de crianças muito prematuras, respondendo aos seguintes objetivos específicos:

1. Identificar as fontes de *stress* de mães e pais de crianças muito pré-termo hospitalizadas em UCIN, e a associação com características sociodemográficas, obstétricas e relacionadas com a criança.
2. Validar o Inventário de Necessidades da Família em UCIN na população portuguesa e propor uma Versão Curta do mesmo.
3. Explorar as necessidades de mães e pais de crianças muito pré-termo hospitalizadas em UCIN de acordo com a sua posição socioeconómica, história obstétrica e as características relacionadas com a criança, através da integração de dados quantitativos e qualitativos.
4. Explorar as perspetivas de mães e pais acerca da sua QdV 4 meses após um parto muito pré-termo, através da integração de dados quantitativos e qualitativos.

Métodos

Estudo observacional e longitudinal, usando metodologias mistas. Com um desenho de investigação multifásico, envolveu a recolha de dados quantitativos e qualitativos em dois momentos diferentes: 1) durante o período de hospitalização da criança, 15 a 22 dias após o parto, utilizando questionários quantitativos individuais; 2) após a alta, aproximadamente 4 meses após o parto, utilizando entrevistas qualitativas semiestruturadas em casal e questionários quantitativos individuais.

Entre 1 de julho de 2013 e 30 de junho de 2014, 120 mães e 91 pais de crianças muito prematuras, hospitalizadas numa das 7 UCIN de nível III da Região de Saúde do Norte de Portugal, foram recrutados sistematicamente (proporção de participação = 96,8%). Os questionários estruturados foram presencialmente administrados às mães e aos pais separadamente, mas em tempos aproximados, por entrevistadores treinados. As características demográficas e socioeconómicas, assim como os dados da história obstétrica, foram auto reportados. Informações acerca de complicações da gravidez, tipo de parto, gravidez múltipla e características neonatais da criança foram obtidos a partir da consulta dos processos clínicos relativos à hospitalização na UCIN. Os dados relativos ao suporte social, às necessidades parentais em UCIN e às fontes de *stress* foram recolhidos através de questionários autoaplicados. Aproximadamente 4 meses após o parto, entre 1 de novembro de 2013 e 30 de novembro de 2014, foram enviados questionários de autoaplicação por correio para 113 famílias que previamente aceitaram participar nesta fase do estudo. Destas, 67 mães e 64 pais preencheram individualmente e devolveram os questionários (proporção de participação = 59,3%). Recolheram-se dados acerca da duração do internamento e sobre o diagnóstico de problemas de saúde na criança, bem como sintomas de ansiedade e depressão dos pais, *stress* parental e perceção da QdV. Realizaram-se entrevistas qualitativas a uma subamostra de 26 casais, entre novembro de 2013 e abril de 2014. Constituiu-se uma amostra intencional, considerando a inclusão de pais de crianças com e sem extremo baixo peso ao nascimento, e heterogénea, com o objetivo de obter a máxima variação de perspetivas e experiências até atingir a saturação temática.

Foi utilizada estatística descritiva e inferencial para analisar os dados quantitativos. Procedeu-se à análise de conteúdo temática dos dados qualitativos, utilizando uma estratégia de triangulação. Os extratos com significado semelhante foram sintetizados em categorias, indutiva e dedutivamente (de acordo com os objetivos de cada artigo), e posteriormente agrupados em temas analíticos.

Resultados

Os pais classificaram a experiência global de hospitalização das crianças em UCIN como mais stressante do que a mediana das restantes subescalas. A "alteração nos papéis parentais" foi considerada a maior fonte de stress tanto pelas mães (Mediana (P25–P75): 4,1 (3,2–4,7)) como pelos pais (Mediana (P25–P75): 3,2 (2,4–4,0)). As mães classificaram como mais stressantes todas as subescalas comparativamente com os pais. Nas mães, a ocorrência de uma gravidez múltipla associou-se a níveis mais baixos de *stress* nas subescalas "alteração nos papéis parentais" ($\beta = -0{,}597$; 95% IC = −1,020 a −0,174) e "stress total" ($\beta = -0{,}603$; 95% IC = −1,052 a −0,153). Nos homens, ter 30 ou mais anos de idade associou-se a menores níveis de *stress* total e em todos os domínios (**ARTIGO II**).

A análise fatorial exploratória do Inventário de Necessidades da Família em UCIN revelou duas dimensões, uma focada nas necessidades dos pais e outra nas necessidades da criança. Para construir a Versão Curta do Inventário, os itens com efeito teto foram eliminados e 22 itens foram submetidos a análise confirmatória, que sustentou a existência de duas dimensões (CFI = 0,925). A Versão Curta apresentou um elevado grau de confiabilidade (alfa ≥ 0,76). Os pais mais velhos e aqueles com menor escolaridade atribuíram, mais frequentemente, maior importância à subescala de necessidades centradas nos pais, enquanto os pais de gémeos revelaram uma tendência para valorizar as necessidades centradas nas crianças (**ARTIGO III**).

As mães valorizaram mais as necessidades de informação do que os pais (Mediana (IIQ): 3,8 (3,6-3,9) vs. 3,7 (3,5-3,9)). Os homens sem outros filhos, assim como as mães mais velhas e com menor escolaridade reportaram sentir mais necessidades do que os restantes participantes. Para além das diferenças de género, as necessidades de confiança nos profissionais e serviços de saúde, assim como de proximidade foram valorizadas por todos os pais, independentemente da UCIN. Os dados qualitativos acrescentaram as seguintes necessidades: suporte instrumental do governo; suporte emocional regular de psicólogos e técnicos de serviço social; melhoria da privacidade na unidade de neonatologia para garantir a prestação de informação e conforto centrados na família; e disponibilidade de outros pais e profissionais de saúde para atuar como mediadores de saúde complementares na prestação de informação detalhada e coerente (**ARTIGO IV**).

Não se observaram diferenças de género significativas na perceção da QdV, registando-se scores semelhantes aos da população portuguesa. Os valores de QdV aumentaram ligeiramente do domínio ambiente (Média (DP): 72,1 (14,2)) para o domínio psicológico (Média (DP): 78,7 (14,4)). O *stress* parental e os sintomas de ansiedade e depressão influenciaram negativamente a QdV de mães e pais. Uma posição socioeconómica baixa influenciou negativamente as perceções de mães e pais no domínio ambiente, assim como a QdV física e psicológica das mães. Os fatores relacionados com a criança (extremo baixo peso ao nascimento, hospitalização em UCIN durante 2 ou mais meses ou problemas de saúde) associaram-se negativamente à QdV geral das mães e aos domínios físico, psicológico, social e ambiental da QdV dos homens. Os resultados qualitativos revelaram mecanismos de acomodação acionados pelos pais para contrabalançar os fatores constrangedores (vigilância, perturbações no sono, falta de suporte das políticas de saúde, higienização) e facilitadores (suporte social, acessibilidade/qualidade dos cuidados de saúde, oportunidades de desenvolvimento de competências parentais) da QdV. Estes processos ancoram-se na centralidade da criança e na construção de hierarquias de esperança e expectativas sobre a saúde e o desenvolvimento da mesma (**ARTIGO V**).

Conclusão

Esta tese gerou conhecimento que poderá servir para sustentar o desenvolvimento de políticas e práticas de saúde integradas e centradas na família em neonatologia, ao analisar a QdV dos pais após o nascimento de uma criança muito prematura. Tendo por base uma abordagem de saúde pública, foram consideradas características individuais, familiares, socioeconómicas e políticas. Os resultados sugerem que os fatores relacionados com as características sociodemográficas e com a história reprodutiva influenciam o *stress* e as necessidades dos pais durante a terceira semana do internamento da criança em UCIN, de forma diferente em mulheres e homens. Os aspetos relacionados com as crianças e com as características psicossociais dos pais associam-se com a QdV parental 4 meses após o nascimento da criança. Os mecanismos individuais de acomodação, bem como os fatores estruturais, influenciam a QdV dos pais ao longo de toda a experiência de parentalidade de uma criança muito pré-termo.

Este estudo propõe a inclusão de cinco tópicos adicionais na conceptualização dos cuidados de saúde integrados e centrados na família: a análise da QdV dos pais como um resultado do modelo; a avaliação das necessidades dos pais e das fontes de *stress* em UCIN, tendo em conta a influência do género; considerar a influência das trajetórias reprodutivas de mães e pais, assim como do grau de privacidade na unidade de neonatologia; contemplar o papel da comunidade e da família alargada; e introduzir o contexto socioeconómico e político em que as famílias vivem (por exemplo, governação, financiamento e recursos em saúde; assistência social; trabalho; normas e valores socioculturais).

Os resultados desta tese sugerem, ainda, a implementação de intervenções focadas na redução do *stress* parental e dos efeitos desempoderadores da vigilância e da higienização na QdV dos pais, com o objetivo de minimizar diferenças na saúde das famílias. Além disso, importa assegurar serviços de saúde integrados através da garantia de suporte instrumental por parte do governo; da disponibilização de suporte emocional regular por parte de psicólogos e técnicos de serviço social; da melhoria da privacidade nas unidades de neonatologia para que a prestação de informação e o conforto se centrem mais na família; e da disponibilidade de outros pais com crianças internadas e dos profissionais de saúde para serem mediadores de saúde complementares na prestação de informação detalhada e coerente. Por fim, esta tese alerta para a necessidade de flexibilidade e sensibilidade na investigação científica para desenvolver instrumentos que avaliem as fontes de *stress*, as necessidades parentais e a QdV dos pais, tendo em conta a importância do género, do suporte social, da posição socioeconómica e das trajetórias reprodutivas dos pais, assim como as questões de privacidade e disponibilidade de suporte emocional regular em UCIN.

Numa perspetiva de saúde pública, esta tese convoca questões que devem ser tidas em conta na coprodução de políticas públicas intersectoriais centradas na família, de serviços de saúde integrados e de intervenções focadas na promoção da QdV dos pais. A abrangência da abordagem adotada nesta tese possibilita a sua aplicabilidade em vários contextos neonatais e contribui para repensar a governação em neonatologia, promovendo a coordenação dos cuidados com e em torno das necessidades das crianças, das suas famílias e das comunidades.

1. Introduction

The introductory chapter of this thesis aims to provide a comprehensive overview of the Quality of Life (QoL) of families with preterm infants. It is argued that further mixed-methods research on mothers' and fathers' experiences, needs, stress and QoL after a preterm childbirth is needed for designing and implementing effective Family Integrated Care (FICare).

The first section covers issues related with the conceptualization and assessment of QoL. Being a holistic and multidimensional concept, the assessment of QoL must take into account the influence of individual and social characteristics and its subjective and objective dimensions. The lack of a quantitative instrument specifically designed to assess the impact of a very preterm childbirth on parental QoL claims for the use of a mixed methods approach to capture such complex experience.

The following section describes the epidemiology of prematurity, and presents a literature review about parental experiences and QoL when caring for preterm infants. An increasing number of epidemiological and public health studies have been focused on the QoL of children and adults born preterm, while parental QoL after a preterm childbirth is clearly understudied both during and after hospitalization period.

The final section explores the missing links in the literature about how parents should be involved in the co-production of health inside and outside Neonatal Intensive Care Units (NICUs). The implementation of FICare entails individual, organizational and political challenges that have been addressed mainly through the perspectives of healthcare professionals. Time is ripe to also listen the point of view of both mothers and fathers.

1.1. Quality of life

1.1.1. Conceptual framework

The recognition of health as a state of complete physical, mental and social well-being, by the World Health Organization (WHO) in 1947 (1), constitutes a landmark in the election of QoL as a privileged research outcome in the field of health sciences. QoL emerged in the medical literature in the 60's and it was introduced in the PubMed database as a keyword in 1975 (2), being currently defined as "person's perception of his/her position in life within the context of the culture and value systems in which he/she lives and in relation to his/her goals, expectations, standards and concerns" (3).

Contemporary thinking about QoL is influenced by old theoretical multidisciplinary debates about what defines the optimal experience, what constitutes the good life or what happiness means (4-6), as well as their objective/subjective and individual/societal nature. During the first half of the 20th century, the societal analysis of QoL has been inspired by a mercantilist approach, according to which the levels of a state welfare (i.e., levels of income, expenditures and savings, the production and sales of goods and services, and commercial activities) determined the QoL of the population (5, 7). However, the absence of a direct association between economic welfare and QoL was consistently observed worldwide (8), claiming for the analysis of other social indicators, such as educational level, type of housing and neighbourhood crime rates (5, 9). The understanding of the influence of social indicators on population QoL was refocused in the 70's by a growing interest on psychological characteristics and individual QoL (5, 9).

During the 90's, two relatively distinct, yet overlapping, dominant approaches to the conceptualization of QoL highlighted its individual and subjective nature: the hedonic perspective stressed the nobility of persons and put emphasis on personal freedom, self-preservation and enhancement, sustaining that QoL consisted on the achievement of happiness and pleasure and avoidance of pain; while the eudaimonic view, based on the Aristotelian conception of "the good life", moderation, reason and justice, considered that QoL consisted in the fulfilment or realization of one's self or true nature (4, 10). These approaches were often contested by their ambiguity and limited applicability (5, 10). One of the most important arguments sustaining this criticism consists in the idea that individuals are not trustworthy subjects to assess their QoL because their views would always be circumstantially built and not based on accurate evaluations. Thus QoL should be assessed through objective measures of feelings associated to the real state of happiness (5, 10, 11). Closely following a utilitarian approach, QoL is seen as related to having objective "good experiences" (5). This conception has also been criticised for neglecting the different meanings attributed by individuals to similar positive events (12), which reinforces the need to look upon QoL as a multidimensional phenomenon including subjective and objective measures (4). Happiness, life satisfaction and subjective well-being are considered building blocks of QoL, but such subjective attributes are not enough to accurately assess individuals' QoL (10).

Nowadays, it is widely recognised the need for a holistic approach to QoL that takes into account the influence of individual and social characteristics as well as its subjective and objective dimensions. This approach evokes an analysis of several interrelated dimensions of life domains, such as individual's physical health, psychological state, level of independence, social relationships, personal beliefs, relationships to the environment, spirituality as well as global physical and social environmental sustainability, social and community resources (e.g., civic integration, synergy and integrity, network links and bridging ties in all levels of the society), and extensive social norms and values (e.g., trust, reciprocity, fairness, equity, social justice and egalitarianism) (10, 13).

In this thesis the analysis will be based on three main theoretical models that are in line with the WHO conceptualization of QoL: the "being, belonging and becoming" model (14); the "human ecological model" (6); and the Hancock's public health model (15). Drawing on the Raphael's "being, belonging and becoming" model (14) we will discuss the degree of enjoyment resulting from opportunities with importance to the person by analysing individual physical, psychological and spiritual characteristics ("being"), his/her adaptation to physical, social and community environments ("belonging"), and the purposeful activities carried out to express oneself and to achieve personal goals and aspirations ("becoming"). The Bulbolz et. al's "human ecological model" (6) will be used to explore the degree of satisfaction with basic physical, biological, psychological, economic and social needs in the microsystem (e.g., material and health conditions, and the significant personal relationships network), the mesosystem (e.g., neighbourhood and community), and the macrosystem (e.g., the culture and health policies) (5, 6). We will also draw upon Hancock's public health model (15) to understand the influence of an adequate and prosperous economy, which fully meets the individuals' basic needs, a viable environment which sustains human life and well-being, and a friendly community including supportive networks, on individuals' QoL.

1.1.2. Assessment

Instruments designed to evaluate QoL can be broadly grouped based on the approach to measurement (objective/directly observed or subjective/self-reported or using a proxy), on specificity (generic or disease/social group-specific), and on the dimensions of QoL they measure (16). The majority of the QoL questionnaires explores domains related to overall or global QoL, physical health/health functioning, emotional or psychological/mental health as well as social relationships and participation in social activities (17-20). There are other dimensions only captured by few instruments, such as material well-being/socioeconomic/money (17, 20, 21), level of independence (19), personal development (17), self-esteem, goals and values, and environment/satisfaction with home, neighborhood and community (21). More recently the spirituality/religion and religious beliefs were also considered an important dimension of QoL (22).

Many early questionnaires used objective measures based on the assessment of dimensions classified as important from the perspective of health professionals (23). One example is the QL-Index, developed by Spitzer (2, 24), a scale where the medical doctor, as an observer, evaluates not only the physical state of patients but also their psychological, social and spiritual dimensions (24). Contrasting with these traditional measures of health and illness, which were the gold standard both in medical and social science fields, several authors currently defend an assessment of QoL based on subjective self-reports (25). Some of the first self-reported questionnaires were based on reviews and adaptations of existing scales designed to measure health status, such as the *Nottingham Health Profile* (26), the *European Quality of Life Measure* (EuroQoL) (27), as well as the *36 item Short-Form Health Survey* (SF-36) (18). In some self-reported instruments, each item is rated in relation to the importance attributed by a person, but the majority weighed them equally assuming that they have the same value for the respondents (28). Self-reported instruments can be administered by a trained interviewer or by the individual. The first mode implies having skilled human resources but ensures compliance, decreases errors and missing data, while the second method of administration is less expensive but potentially increases the number of missing data. Thus, the best approach may be a compromise - having participants filling in the questionnaire supervised by a researcher (28).

Another common measurement approach is using a surrogate respondent to predict the results that would be obtained by a patient or children (28). Proxy respondents are usually selected on the basis of intimate knowledge of the individual, or on their professional skills to make judgements on another person’s behalf (for instance, a family member, a caregiver or the medical doctor). However, results across studies show that scores by different types of proxy may not be equivalent or interchangeable and can be very different from those self-reported (29-31).

Some studies showed that family and nurses proxies tend to underestimate individuals' QoL (29, 32), while others revealed that clinicians tend to overestimate QoL in comparison with patients' self-report (33). Previous research assessing the agreement between parental and children reports of QoL demonstrated some agreement on physical domains, but they showed discrepancies related to more subjective domains such as social and emotional functioning (34, 35). Thus, studies using surrogates for QoL assessment should carefully discuss their results.

Generic instruments are intended to address the most common areas of QoL for the general population. They are applicable to all persons independently of their type or number of illness, and they are very useful when the purpose of the study is the comparison between groups or individuals with different health conditions or to measure treatments' complications not directly related to the disease. Nevertheless, they can omit some domains that are important to a specific group of people or disease conditions, hindering their ability to detect clinically important changes following treatment or interventions (36, 37). The most widely used generic instrument is the *World Health Organization Quality of Life inventory* (3). Representing an international and transcultural effort to standardise the assessment of QoL, this instrument allows a collaborative research in different cultural settings and the direct comparison of results (19). However, the WHO recognises that there may be some aspects of QoL which are important in a culture but not covered by the instrument, admitting the possibility to add modules to the core instrument in order to assess people with a particular disease or in circumstances in which the core modules do not provide sufficient detail (3).

A few instruments have been developed to assess specific diseases (e.g., heart failure), populations (e.g., parents), functions (e.g., sleep) or problems (e.g., pain) (28). They are more sensitive but their results are difficult to interpret in individuals with multiple diseases as well as difficult to compare between different groups of people (37). In the last decades, questionnaires aiming to assess the effect of caring for infants with specific health conditions on caregivers' QoL (38) have been developed, but none of them was designed for parents of preterm infants. Considering that generic questionnaires used to assess QoL of parents of sick children may not be sensitive to differences relevant to the child's condition or treatment (38, 39), specific questionnaires are invaluable in estimating the parental burden and types of healthcare, psychological and social services parents may need (38). Qualitative studies are needed to identify and to understand the factors valued by people present and absent from the questionnaires and their relative importance (23). Thus, to have a more complete knowledge on QoL and to capture the complexity of its assessment, researchers and clinicians have been defending the use of mixed methods approaches to moving forward in uncovering the pathways that lead to people judgments (36, 40).

In addition, researchers tend to create short-versions of the original QoL instruments, brief and easy to administrate in order to diminish the individuals' burden associated with assessment and increase their use in clinical practice and large-scale epidemiological studies. Those short-versions usually tend to decrease the number of items included in the questionnaire while maintaining the most significant domains to measure (41).

1.2. Prematurity

1.2.1. Epidemiology

Preterm birth occurs before 37 completed gestational weeks or 259 days of gestation (42). The negative influence of preterm birth on individuals, families and societies, as well as the healthcare costs associated with perinatal care and long-term disability (43, 44), make preterm birth a major public health priority (45), with preterm infants being at high risk of neonatal morbidity and its complications constituting one of the leading causes of global deaths among children under 5 years of age (46, 47).

According to gestational age, a preterm infant could be classified as late preterm (34-36 weeks of gestation), moderate preterm (32-33 weeks of gestation), very preterm (28 to 31 weeks of gestation) or extremely preterm (<28 weeks of gestation) (48). Although some studies have been used interchangeably the concepts of gestational age and birth weight, they should be distinguished because there is a range of a normal or expected birth weight per gestational age and sex. In fact, a baby born preterm has a higher risk of death than a baby born at term with the same birthweight, even being small for gestational age (49, 50).

Preterm birth could be iatrogenic when medically indicated or spontaneous due to spontaneous preterm labour and/or premature rupture of membranes (45, 51). The aetiology of spontaneous preterm birth is complex and multifactorial, and its causes remain unknown in many cases. The literature demonstrates an association between preterm birth and several genetic, maternal and foetal characteristics, as well as environmental factors, including individual or family history of preterm birth, maternal sociodemographic characteristics (e.g., young or advanced maternal age, black women), nutritional status (e.g., low prepregnancy BMI), pregnancy history (e.g., short interpregnancy intervals), current pregnancy characteristics (e.g., multiple pregnancy), psychological characteristics, adverse behaviours (e.g., working long hours and undertaking hard physical labour under stressful conditions), infection, uterine contractions and cervical length (45, 50, 52, 53). However, several studies show that spontaneous preterm births also occur in women without identifiable risk factors (51).

Recently, it has been proposed that successful prevention of preterm birth requires a multifaceted approach, combining public health and educational interventions, lifestyle modification, the optimisation of obstetric care, and the application of effective, targeted interventions (54). However, current strategies to prevent preterm birth focus largely on managing risk factors, namely maternal smoking cessation, progesterone treatment, decreasing multiple embryo transfers during Assisted Reproductive Technologies (ART), cervical cerclage as well as reduction of non-medically indicated labour induction or caesarean section (55, 56).

Globally, the average preterm birth rate in 2010 was estimated at 11.1% (14.9 million infants), corresponding to more than one in ten of all births. Preterm rates varied widely across countries, ranging from about 5% in several northern European countries to 18% in Malawi. Even though rates are highest for low-income countries, high preterm birth rates were also described in many high-income countries (e.g., 12.0% in the USA) (50) (Figure 1). In Europe, relatively lower preterm birth rates (<6.5%) were observed in Iceland, Lithuania, Finland, Estonia, Ireland, Latvia, Sweden, Norway, and Denmark, contrasting with the highest rates in Hungary (8.9%) and Cyprus (10.4%) (57).

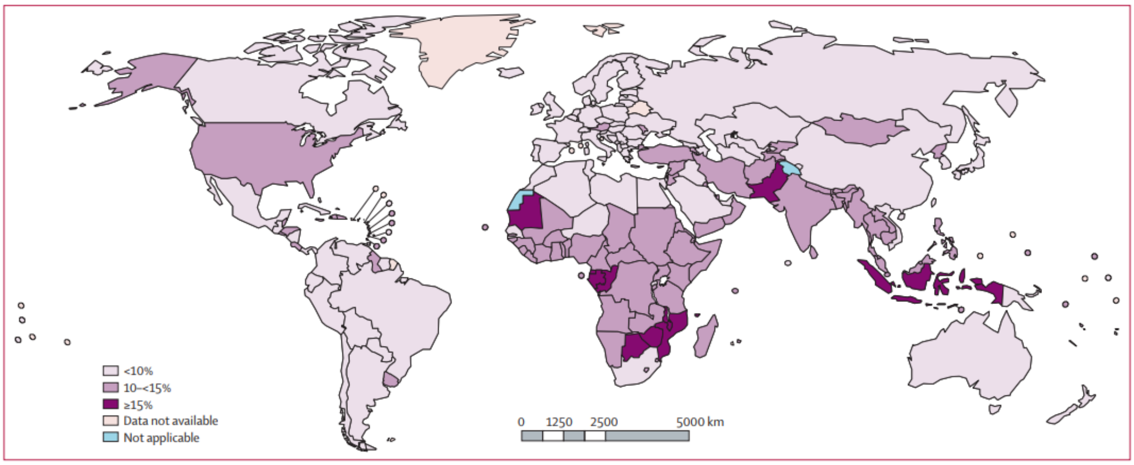

Figure 1. Worldwide estimated preterm birth rates by country for 2010 (50)

The estimation time trends for 65 countries in Developed, Latin America and the Caribbean regions demonstrated that in almost all countries the preterm birth rate has increased from 1990 to 2010 (50). Possible reasons contributing for this trend include the increasing maternal age and higher maternal body mass index, the rise of multiple births associated and non-associated with the use of ART, and the non-medically indicated inductions and caesarean sections (56). Preterm birth rates also rose in most of the European countries from 1996 to 2008, mostly due to the rises in the multiple birth rates and the preterm birth rate for multiple births (58). In Portugal they increased 0.9% between 2004 and 2010, representing 7.6% of the total live births in 2010 (57). The last national statistics showed that the preterm birth rate in Portugal is still increasing, being 8.1% in 2017 (59).

Very preterm birth, occurring before 32 gestational weeks, accounts for about 1% of worldwide live births and represents a proportion of about 16% of all preterm births (48). In Europe, the rates varied from 0.7% for Iceland to 1.4% for Brussels (Belgium) and Hungary in 2010 (Figure 2) (57). In Portugal, 912 very preterm babies were born in 2017, representing about 1.1% of all births and a proportion of approximately 13% of preterm births (59).

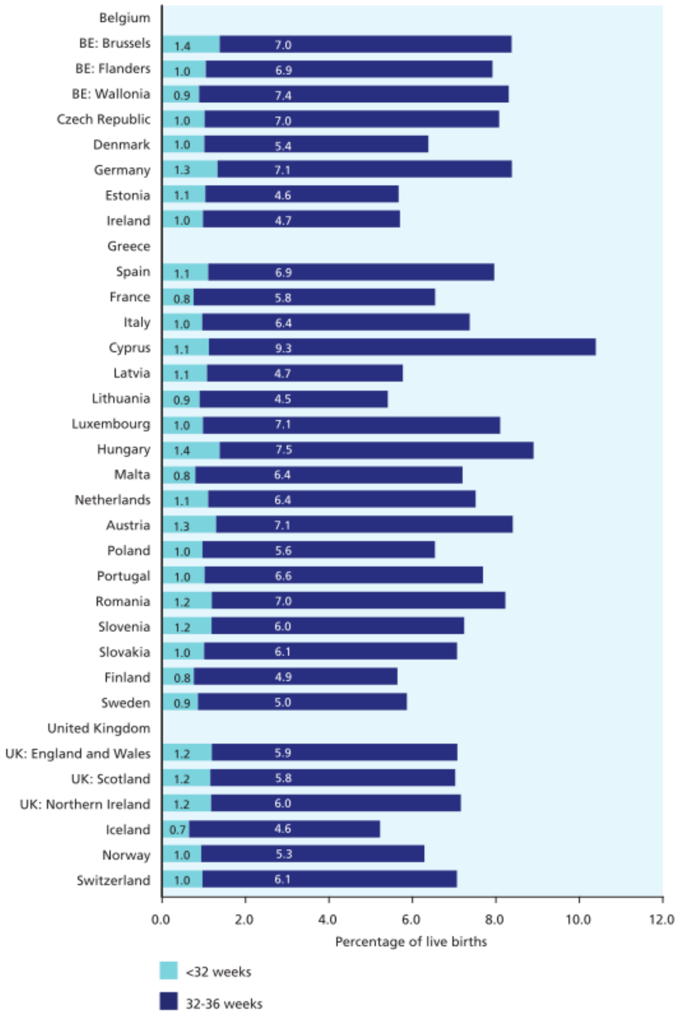

Figure 2. Percentage of live births with a gestational age <32 weeks and between 32-36 weeks in European countries, 2010 (57)

More than 90% of very preterm babies born in Portugal were delivered at level III public hospitals that are referral centres with differentiated perinatal support, caring for high-risk pregnancies and infants with obstetricians, neonatologists and NICUs. Portuguese NICUs have between five and 12 intensive care cots and provide long-term ventilation, high frequency oscillation, inhaled nitric oxide and early nasal continuous airway pressure (57, 60). There are seventeen differentiated perinatal support hospitals in Portugal, seven of them located in the northern health region (61). Very preterm infants are highly dependent on intensive care after birth mainly due to neonatal complications reflecting the immaturity of organ systems, which implies a hospitalisation in a NICU. This intensive care manages feeding and growth difficulties, constant monitoring and early treatment of neonatal problems such as breathing difficulties, infections, and jaundice (47, 50), but there is a wide difference in the care provided to these infants across Europe. A multinational prospective observational population-based study (EPICE project), analysing 19 regions from 11 European countries, found that only 58.3% of very preterm infants admitted to a neonatal unit received the appropriate evidence-based care (e.g., delivery in a maternity unit with appropriate level of neonatal care; administration of antenatal corticosteroids before delivery; prevention of hypothermia; surfactant used within two hours after birth or early nasal continuous positive airway pressure) (62). These disparities in care may increase the inequities among very preterm infants with an impact on their short and long-term health outcomes as well as on family burden. How these disparities affect the QoL of families and are influenced by their socioeconomic status is an important area for future studies (63).

The increasing survival chances reported over the last three decades as well as the high prevalence of health and developmental problems of preterm infants led to a rising concern about the influence of preterm birth on QoL of individuals born preterm across the life course (34, 47, 62, 64), while data regarding the QoL of their mothers and fathers during and after hospitalisation in a NICU is scarce. Very preterm infants are at an increased risk of developing physical (e.g., respiratory, gastrointestinal and visual impairments), neurodevelopmental (e.g., cerebral palsy) and behavioural (e.g., attention deficit hyperactivity disorder) problems (47, 65, 66). Moreover, these infants need an intensive follow-up especially during the first years of life, having more readmissions and almost 10 times more outpatient visits than full-term children, resulting in a high burden for their parents and families (47, 64, 67). Several studies have also demonstrated that the infants born preterm have poorer cognitive, physical, emotional and social functioning than infants born at term during preschool age and adolescence (68, 69) but the majority do not perceive their QoL as significantly different as others of their own age (70). Considering the burden associated with caregiving a very preterm infant, there is a need for future studies exploring the influence of such experience on parental QoL. This knowledge is key for designing, developing and implementing appropriate and effective interventions to tackle parental needs, improve mental health and QoL, with benefits for parents, children and their families (71-73).

1.2.2. Parental roles and experiences

Studies surrounding parental roles and experiences when caring for preterm infants covered four main areas: parenting stress, parental needs, emotions, and coping strategies (74). Mothers and fathers with preterm infants hospitalised in NICUs face multiple stressors that may interfere with the parent-child relationship. A particularly intense effort to establish their roles as parents is required, due to the physical separation from the child, structured and controlled opportunities for interaction, difficulties in participating in the infant's care, and the fear and uncertainty about infant's survival and future health and development (75-77). In addition, the complexity of a NICU environment, with unknown specific smells and lights, noisy life support and monitoring equipment, lack of privacy, and the constant presence of healthcare professionals, has been described as a major contributor to parents' distress (78-81). Contrary to what happens in a regular nursery, where care is provided by parents since birth, parents in NICUs remain consigned to a supportive role, with some of them describing themselves as simple voyeurs, only allowed to visit and hold their infant (82, 83), and feeling anxious with the discharge moment by not being prepared for assuming the care at home (84). Parents need to redefine and adapt their expectations about parenting roles when dealing with the loss of an expected healthy baby and the "phantasy self-as-mother" due to their incapacity to keep the baby alive without medical intervention, decreasing their parental confidence (85-87) and challenging the norms associated with the conventional parenthood (88). The literature shows that parenting stress in NICUs is influenced by a variety of sociodemographic, obstetric and infant characteristics, including parental age, ethnicity, socioeconomic status and exposure to other stressful life events, as well as pregnancy planning and previous pregnancy loss, maternal mental health, severity of infant illness and previous experience of infant's hospitalisation in a NICU (89-91). However, the great majority of these studies are not specifically focused on sources of stress among parents of very preterm infants, were mainly conducted in the USA and tended to exclude fathers, offering a one-sided perspective that fails to approach the influence of the NICU hospitalisation on parents and families. The assessment of sources of stress during very preterm infants' hospitalisation in a NICU and its associated factors is thus essential to identify mothers and fathers at risk for immediate and extended physical and emotional burden.

In settings characterized by high levels of dependence on health professionals such as NICUs (92, 93), mothers and fathers of hospitalised infants highlight their needs for clear, constant and reliable information, as well as for guidance about the care adapted to the infant's illness trajectory phase and the cultural background of the family in order to increase their sense of control (94-98). Parents need to trust in healthcare professionals, by recognizing their technical skills, receiving consistent and continuous care, establishing genuine relationships by receiving reassurance, encouragement and praise and by feeling involved in decision-making processes and caregiving activities (98-102). Parents also recognise their needs for emotional, instrumental and religious support provided by spouses, family, friends, peers and healthcare professionals (95, 102-105). Literature also reported some comfort needs, such as having a comfortable waiting room, vending machines in the waiting areas and lockers for all parents in NICU (103, 106). Parents' needs are likely to vary across their socioeconomic characteristics (e.g., gender, age, marital status, race, educational level, income), cultural expectations as well as the existence of previous children, previous experience in NICU, infant's illness trajectory and length of stay, and the specificities of each NICU (97, 99, 100, 105, 107-110). In addition, these needs seem to be differently perceived by parents and healthcare professionals (106, 111), which could lead to a gap between the care expected by parents and the care actually provided (112). Hence, a deep understanding of parental needs may be useful to the design of strategies aiming to fulfil such needs, contributing to the reduction of the stress related to parenting a hospitalised infant as well as to the improvement of parental QoL (95).

Moreover, parents deal with mixed emotions, where feelings of self-blame, guilt, lack of control and helplessness coexist with hope, love and joy (113). Parents tend to cope with the situation by assuming the responsibility for the infant, not only by actively monitoring the infant's medical condition, comforting, diapering, breastfeeding but also passively by deriving self-comfort from being near the baby, developing an emotional attachment and making the environment more homelike (75).

The birth of a very preterm infant is a long-lasting disruptive event, due to parental concerns about infant's health and development which are often not confined to the postpartum period (114). As pointed out previously, parents often must manage numerous medical and developmental needs beyond those required for a healthy full-term infant, for many months or even years after the NICU discharge, such as re-hospitalisations, several medical appointments, and treatments (115, 116). Caring for a very preterm infant requires an intense care and vigilance which affect parents' QoL in several ways. Research has shown that the first year of age is particularly burdensome for these families (115), with mothers and fathers reporting higher levels of anxiety and depressive symptoms than full-term mothers throughout the first year of infant's age (76, 117). Parents may find the post-discharge period a stressful extension of their NICU stay, not only due to concerns about their infant's safety at home without continuous medical monitoring and their ongoing development but also due to other factors not related with infant's health and sociodemographic characteristics such as social isolation, marital distress, financial burden (e.g., costly medical bills and non-medical expenses related to the loss of work), and an unsafe home environment (118-121).

These findings highlight the complexity of a multifaceted parental experience and point out the importance of recognizing emotional responses at the individual level, taking into account the cognitive appraisals and social and structural factors affecting mothers' and fathers' QoL after a preterm childbirth (74). When facing a catalyst event such as a preterm childbirth, parents may enact behavioural, cognitive and affective processes necessary for accommodating such event in their lives. These accommodation mechanisms (e.g., social comparison, social support, reordering of goals, reframing expectations, spiritual practice) can change parents' internal standards, values or conceptualization of QoL. Considering that the meanings attributed to QoL can change over time as a result from the way individuals deal with stressful life events (23, 25, 122), the "response shift" should be considered when studying the QoL of parents of preterm infants (31, 123). A better understanding of the influence of a preterm childbirth on parental QoL, as well as on modifiable factors that affect it, is essential for designing and implementing family integrated care policies and practices, during and after hospitalisation in a NICU (120, 124-127). Thus, a scoping review, aiming to synthesize the body of knowledge on the factors influencing the QoL of mothers and fathers of preterm infants, was performed and will be presented in the next section.

1.2.3. PAPER I: Quality of life among parents of preterm infants: A scoping review

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REVIEW

Quality of life among parents of preterm infants: a scoping review

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Abstract

Purpose To synthesize the body of knowledge on the factors influencing the QoL of mothers and fathers of preterm infants.

Methods A scoping review was performed. Publications indexed in PubMed®, Web of Science™, CINAHL® and PsycINFO® were searched, targeting studies presenting original empirical data that examined parental perception on QoL after a preterm delivery. Eligibility and data extraction were conducted by two independent researchers. The main quantitative findings were synthesized and qualitative data were explored by content analysis.

Results The studies, 11 quantitative and 1 mixed methods, were derived mainly from the USA ($n=6$). Heterogeneity across the studies was observed regarding the operationalization of QoL and the use of units of analysis (mothers, parents, families and caregivers). In a context where 40 out of 45 covariates were analysed by only one or two studies, results suggested that parental QoL after a preterm delivery is influenced by factors related with mother's characteristics, family issues and health care environment rather than infants' variables. Factors regarding fathers' characteristics and structural levels were not addressed.

Conclusions Standardizing the operationalization of the QoL when analysing mothers and fathers of preterm infants calls for a structured questionnaire adapted to their specific needs. Further research should include both mothers and fathers, invest in mixed methods approaches and be performed in different countries and settings for allowing integration and comparison of findings.

Keywords Quality of life · Premature birth · Parents · Family-integrated care · Scoping review

Introduction

Preterm birth, occurring before completing 37 gestational weeks [1], constitutes the leading cause of neonatal mortality and morbidity worldwide [2]. The increased preterm birth rates and the higher survival chances of these infants, consistently reported over the last three decades [3], have justified the increasing number of studies concerning health and quality of life (QoL) of children and adults born preterm [4–6]. These studies aim to develop optimal evidence-based effective perinatal intensive care [7] and to standardize approaches to the organization of care and medical interventions [8]. Recent studies point to the need to also acknowledge parents' experiences and views focused on developing family-integrated neonatal services [9, 10]. Nevertheless, the factors influencing parental QoL during and after preterm infants' hospitalization in Neonatal Intensive Care Unit (NICU) have not been systematized.

A preterm birth affects the family environment not only during the infant's hospitalization in NICU, but for many years [11]. Mothers and fathers of preterm infants revealed an increased risk of developing parental stress [12–14], depressive symptoms and anxiety shortly after delivery [15, 16], as well as poorer family functioning and higher family burden several years after birth, when compared with families of full-term infants [17]. Existing literature reviews provide information about parental mental health

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and stress, and identify the effects of a preterm delivery on the family structure, with a specific focus on parents of very or extremely low birth weight infants [18], with or without neuropsychomotor disturbance [19]. These reviews were not performed with a systematic methodology neither focused on the factors influencing QoL of mothers and fathers of preterm infants, defined as the individuals' perception of their own physical, psychological, social and environmental well-being, taking into account their culture and value systems, goals and expectations [20]. Such systematic knowledge could represent a relevant tool for designing and developing sustainable and effective family-centred and integrated health care when parenting a preterm infant. Moreover, the evidence provided would contribute to enrich medical practices thus improving health governance in the context of prematurity.

This scoping review aims to synthesize the body of knowledge on the factors influencing the QoL of mothers and fathers of preterm infants.

Methods

We followed the guidance for descriptive systematic scoping reviews by Levac et al. [21], based on the methodological framework developed by Arksey and O'Malley [22].

Stage 1: Identifying the research question

The central question guiding this scoping review is the following one: What are the main factors influencing the QoL of mothers and fathers of preterm infants?

Stage 2: Identifying relevant studies

A search of the publications on four electronic databases (PubMed®, Web of Science™, CINAHL® and PsycINFO®) was undertaken in July 2017, with no restriction set for language or time of publication, using the following search expression: ("QoL" OR "quality of life" OR "life quality" OR "life qualities") AND ("mother" OR "father" OR "mothers" OR "fathers" OR "parent" OR "parents" OR "family" OR "families" OR "maternal" OR "paternal" OR "parental") AND ("birth, premature" OR "births, premature" OR "premature births" OR "preterm birth" OR "birth, preterm" OR "births, preterm" OR "preterm births" OR "infants, premature" OR "premature infant" OR "preterm infants" OR "infant, preterm" OR "infants, preterm" OR "preterm infant" OR "premature infants" OR "neonatal prematurity" OR "prematurity, neonatal"). The search was followed by backward reference tracking, examining the references of the selected publications based on full-text assessment.

Stage 3: Study selection

The inclusion criteria allowed only empirical, peer-reviewed, original full-length studies that explored the QoL of mothers and/or fathers of preterm infants as their main outcome. The exclusion criteria disallowed studies focusing on the QoL of infants, adolescents or adults born prematurely, studies with data about parents' QoL only during pregnancy and studies in which the infants' gestational age was above 37 weeks.

The first and the last authors (M.A. and E.A.) independently screened all the papers retrieved initially, based on the title and abstract and afterwards, based on the full-text. This was crosschecked in both phases. The study selection was guided by the research question and inclusion and exclusion criteria. An almost perfect strength of agreement was achieved in both phases (total percentage of agreement = 96.5%; Cohen's kappa = 0.91, 95% CI 0.87–0.96). Disagreement was solved by joint discussion until consensus could be reached or, when consensus was not achieved, by the assessment of the second author (S.S.), based on the same criteria defined for study selection.

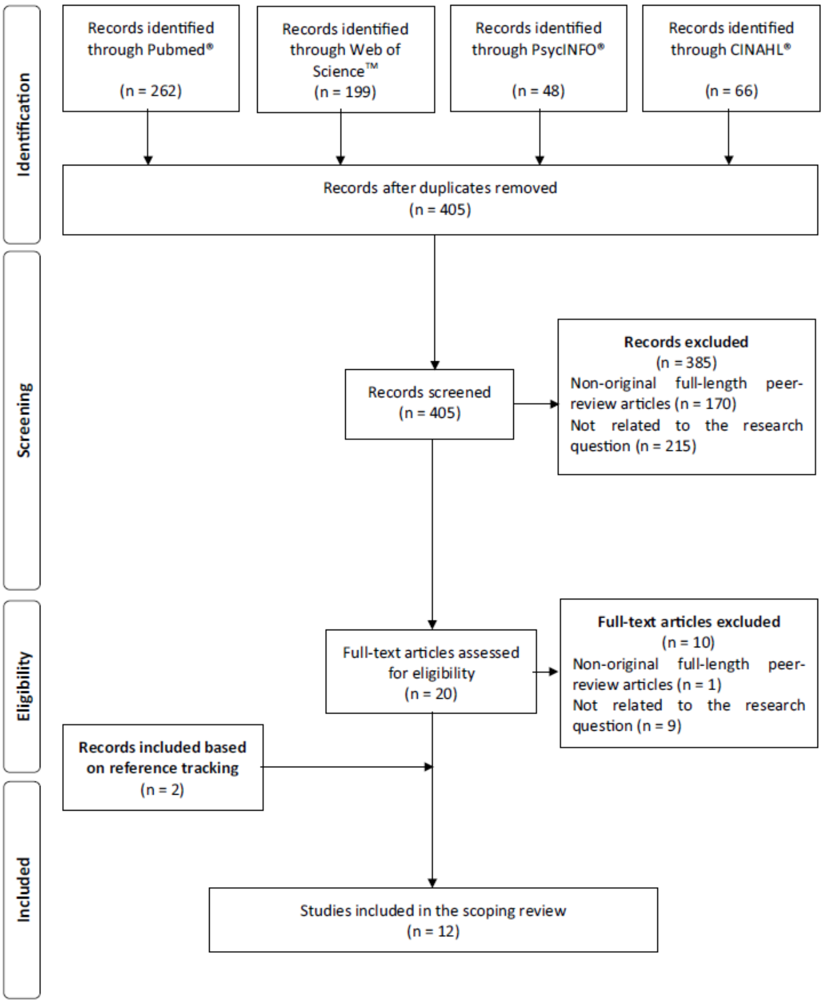
The screening process is summarized in Fig. 1. The titles of 575 records were retrieved. After the removal of the duplicates, 405 records were examined. Based on title and abstract assessments, 385 records were excluded, mainly because they were neither original full-length peer-review studies nor explored parental QoL as the main outcome. Of the 20 fully read papers, 10 met the inclusion criteria. After the backward reference tracking, two papers were included and the final scoping review was composed by 12 papers.

Stage 4: Charting the data

A standardized data extraction sheet was developed and completed by two independent researchers (M.A. and E.A.). Descriptive data for the characterization of studies included research design; information about the authors and publication year; country where the study was developed; study aim; participants and sample; and instruments used to assess parental QoL.

We retrieved quantitative data on variables whose association with parental QoL was statistically significant and the directions of the associations were registered. All the remaining variables whose association with QoL of parents of preterm infants was tested and reported were also extracted.

Fig. 1 Flowchart showing the search results and screening process for the scoping review on QoL of parents of preterm infants



The qualitative data presented in only one study [23] were analysed according to the protocol for content analysis developed by Stemler [24]. In each of the main themes identified by the authors of the above-mentioned study, we selected the categories with the highest and the lowest difference between the two interviewed groups (families of infants with cerebral palsy or hydrocephalus and families of neurologically normal infants). Additionally, the most frequently reported categories related with positive and negative impacts of a preterm birth on family QoL were retrieved.

Stage 5: Collating, summarizing and reporting the results

The main characteristics of the 12 studies included can be found in Table 1. Studies were grouped by research design and ordered by the year of publication.

The factors influencing the QoL of mothers and fathers of preterm infants were identified and then grouped into issues related with mother, infant, family and health care (Table 2). The main findings are presented in Table 3.

Table 1 Empirical studies examining QoL of parents of preterm infants ($n=12$)

Publication	Country	Aim	Participants and sample	Assessment of QoL
Cross-sectional studies				
Rivers et al. [23]	USA	Explore the QoL of VLBW infants and their families, by comparing families of infants with neurologic *sequelae* with families of infants who were neurologic intact, 3 to 7 years after the infants' admission to NICU	22 families of neurologically abnormal PT infants admitted to the NICU 15 families of neurologically normal PT infants	Open-ended questions
Donohue et al. [25]	USA	Compare the QoL of caregivers of VLBW infants and of full-term infants, 12 to 18 months after delivery	83 primary caregivers (80 mothers) of VLBW PT infants who had been cared for in an urban inner-city NICU 84 primary caregivers (83 mothers) of full-term infants cared in the well-baby nursery, paired for maternal race, age and parity	Quality of Life Inventory[a]
Lee and Kimble [32]	USA	Explore relationships between impaired sleep and well-being, in mothers of low birth weight infants hospitalized in the NICU, at 2nd week postpartum	20 first-time mothers of LBW PT infants	The 36-item Medical Outcomes Short Form-36 version 2 (SF36v2)[b]
Lee et al. [33]	USA	Describe maternal daytime activity levels and their association with HRQOL, at 2nd week postpartum	51 healthy first-time mothers of LBW PT infants	The 36-item Medical Outcomes Short Form-36 version 2 (SF36v2)[b]
Lee and Hsu [34]	USA	Assess the role of sleep on the association between maternal stress and health-related well-being (depression, fatigue and HRQOL) among mothers with PT infants in the NICU, at 2nd week postpartum	55 first-time mothers of LBW PT infants	The 36-item Medical Outcomes Short Form-36 version 2 (SF36v2)[b]
McGowan et al. [29]	Ireland	Explore the health outcomes and family functioning of late PT infants according to the provision of neonatal intensive care, 3 years after delivery	Mothers of 101 late PT infants (34+0 to 36+6 weeks of gestation) who received any episode of neonatal intensive care Mothers of 122 late PT infants who did not receive any intensive care or who received special care only for up to a maximum of 3 days	The PedsQL Family Impact Module[c]
Case-control studies				
McLean et al. [31]	Australia	Assess the impact of caring for a PT infant receiving HOT on mothers QoL after discharge	10 mothers of PT infants diagnosed with chronic lung disease receiving HOT 10 mothers of PT infants who previously received HOT 20 mothers of PT infants who did not require HOT	The 36-item Medical Outcomes Short Form (SF36)[d]
Sharma and Sohi [26]	NR	Compare self-perceptions of QoL of mothers of PT and term babies during the first 6 months of infant's age	50 mothers of PT babies 50 mothers of term babies	The WHOQOL-BREF questionnaire[e]

Table 1 (continued)

Publication	Country	Aim	Participants and sample	Assessment of QoL
Longitudinal studies				
Hill and Aldag [27]	USA	Examine PT, near-term and term mothers' self-reported QoL at 1st and 3rd weeks postpartum	37 mothers of PT infants (24 to 33 weeks of gestation) 59 mothers of near-term infants (34 to 37 weeks of gestation) 88 mothers of term infants (38 to 41 weeks of gestation)	Maternal Postpartum Quality of Life Instrument (MAPP-QOL)[f]
Mautner et al. [28]	NR	Explore the influence of PT delivery as risk factor for HRQOL between the 24th and 37th weeks of gestation and the 3rd to 4th months postpartum (using three time points)[g]	32 women with a PT delivery 18 women with gestational hypertensive disorders 11 women with gestational diabetes 29 women with uncomplicated pregnancies	The WHOQOL-BREF questionnaire[e]
Nordheim et al. [30]	Norway	Examine parental QoL after participation of their VLBW infants in a nutrition RCT during NICU hospitalization and at 3.5 years of infants' age	31 parents (≈45% fathers) of VLBW PT infants participating in a nutrition RCT 31 (≈50% fathers) parents of non-participating VLBW PT infants	Quality of Life Scale-Norwegian version (QoLS-N)[h]
Moura et al. [35]	Brazil	Identify factors associated with the QoL of mothers of VLBW PT infants during the first 3 years after delivery (using five time points)	75 mothers of PT infants (≤ 34 weeks of gestation) with VLBW	The WHOQOL-BREF questionnaire[e]

HOT home oxygen therapy, *HRQOL* health-related quality of life, *LBW* low birth weight, *NICU* neonatal intensive care unit, *NR* not reported, *PT* preterm (<37 weeks of gestation, except if otherwise specified), *QoL* quality of life, *RCT* clinical randomized intervention trial, *USA* United States of America, *VLBW* very low birth weight (<1500 g)

[a] Domains: health; self-esteem; goals and values; creativity; learning; money; work; play; helping; love; relationship with friends, family and children; satisfaction with home, neighbourhood and community; overall QoL (range – 6 –6)

[b] Domains: physical health; emotional health (range 0–100)

[c] Domains: Global HRQOL (range 0–100)

[d] Domains: vitality; general mental health; role limitations due to emotional problems; role limitations due to physical problems; social function; bodily pain; general health perceptions; physical functioning (range 0–100)

[e] Domains: global QoL; physical health; psychological health; social relationships; environment (range 0–100)

[f] Domains: overall QoL; psychological/baby; socioeconomic; relational/spouse-partner; relational/family friends; health and functioning (range 0–30)

[g] For the purposes of this scoping review, we only used data from the last moment of data collection

[h] Domains: overall QoL (range 16–112)

Table 2 Synthesis of the factors whose association with QoL of parents of preterm infants was tested

Factors (number of studies)	Negative influence	Positive influence	No association
Mother (*n* = 8)			
Mental health	✓✓✓✓[25, 33–35]		✓[35]
Disturbed sleep	✓✓✓[32–34]		
Fatigue	✓✓[33,34]		
Stress	✓[34]		
Pregnancy complications	✓[28]		✓[35]
Time postpartum		✓[27]	
Light exposure		✓[32]	
Religion		✓[35]	
Circadian activity rhythms			✓✓[32,33]
Parity			✓✓[26,35]
Number of miscarriages			✓[35]
Night-time total sleep time			✓[32]
Self-reported physical health			✓[25]
Age			✓[35]
Skin colour			✓[35]
Educational level			✓[35]
Occupation			✓[35]
Working status			✓[26]
Infant (*n* = 7)			
Health problems	✓✓✓[23a,31,35]		✓✓[23b,25]
Gestational age	✓✓[27,35]		✓✓[26.31]
Birth weight		✓[25]	✓✓✓[26,31,35]
Gender		✓[35]	✓[26]
Length of stay in NICU			✓✓[29,35]
Chronological age			✓✓[31,35]
Intrauterine growth restriction			✓[35]
APGAR score at 5 min			✓[35]
Motor and language quotient			✓[25]
Family (*n* = 5)			
Lack/existence of support system	✓[23]	✓[23]	
Memories of neonatal period (Guilt/Optimism)	✓[23]	✓[23]	
Relationships to child (spoiled/high appreciation)	✓[23]	✓[23]	
Presence/absence of impact on parents' lives	✓[23]	✓[23]	
Changed/not changed plans for future children	✓[23]	✓[23]	
Family income	✓[35]		
Stable marital union		✓[35]	
Family set up			✓[26]
Number of adults in home			✓[25]
Place of residence			✓[31]
Health care (*n* = 4)			
Absence/presence of medical information	✓[23]	✓[23]	
Presence/absence of financial problems	✓[23]	✓[23]	
Misbehaviour of medical personnel	✓[23]		
Hospitalization in NICU	✓[35]		
Participation in a nutrition RCT		✓[30c]	✓[30d]
Mode of delivery			✓✓[28,35]
Number of prenatal care visits			✓[35]
Use of antenatal corticosteroids			✓[35]

Table 2 (continued)

NICU neonatal intensive care unit, *RCT* clinical randomized intervention trial

✓Represents the number of studies reporting the respective association

[a] Regarding the dimensions: stress related to the unwillingness of the paediatrician to agree that child had a problem or needed referral for therapy, and difficulties related to the physical work to care for child

[b] Regarding the dimensions: absence of problems with the neonatal hospital bill and the importance given to the support provided by extended family

[c] During infant's hospitalization in NICU

[d] At 3.5 years after delivery

Results

Study characteristics

Research design

Six studies were cross-sectional, two were case–control and four were longitudinal studies. Eight of the studies had at least one comparison group, such as caregivers and mothers of full-term infants [25–27], women with other pregnancy complication than a preterm delivery [28], mothers of near-term (34–37 weeks of gestation) infants [27], parents of very low birth weight (VLBW) infants not participating in a Clinical Randomized Intervention Trial [30], mothers of preterm infants who did not require and previously received home oxygen therapy [31], mothers of infants who did not receive any intensive care or who received special care only for up to a maximum of 3 days [29] and families of neurologically normal infants [23]. The remaining four studies [32–35] assessed the QoL of mothers of preterm infants.

Almost all studies used a quantitative methodology ($n = 11$), assessing QoL through seven different standard instruments. The only mixed methods study relied on interviews using a structured questionnaire and open-ended questions [23]. The timing of data collection ranged from the first to third weeks postpartum during NICU hospitalization [27], until 3 months [28] to 7 years after delivery [23].

Country of study origin and year of publication

Half of the studies were conducted in the USA, and the remaining derived from 4 countries: Ireland ($n = 1$), Australia ($n = 1$), Norway ($n = 1$) and Brazil ($n = 1$). Two studies did not report the country of study origin, being authored by researchers from India [26] and Austria [28]. The studies were published between 1987 [23] and 2017 [35].

Participants and sample

In the total of the 12 papers, samples were composed mostly of mothers ($n = 9$), followed by samples of parents ($n = 1$), families ($n = 1$) and primary caregivers ($n = 1$). In the latter, 97.6% were mothers [25]. The gender of the participants is not specified in the study analysing families [23]. Samples varied from 20 first-time mothers [32] to 223 mothers of late preterm infants [29], and there were 167 caregivers [25], 37 families [23] and 62 parents of VLBW infants [30].

Assessment of QoL

Half of the studies used the WHOQOL-BREF Questionnaire [26, 28, 35] or the 36-item Medical Outcomes Short Form-36 version 2 [32–34]. The remaining quantitative studies assessed parental QoL through the following instruments: The 36-item Medical Outcomes Short Form (SF36) [31]; Quality of Life Inventory [25]; The PedsQL Family Impact Module [29]; Maternal Postpartum Quality of Life Instrument (MAPP-QOL) [27] and Quality of Life Scale-Norwegian version (QoLS-N) [30]. Five instruments assessed the global/overall QoL [25–30, 35], and three studies measured the domains physical health and emotional health using the same instrument [32–34]. The other 29 QoL domains were evaluated by only one study each [25, 27, 31]. Study by Rivers, Caron and Hack [23] evaluated QoL through the following questions: How has your life been changed by the birth of your premature child?; What has the financial impact of your child's birth been on your family?; How has your child's birth affected your plans for future children?

Factors influencing the QoL of parents of preterm infants

Factors related with mothers and infants' characteristics were more frequently addressed, followed by those centred on the family and health care. Across all the studies assessed, 45 variables potentially associated with QoL were identified, and in most studies few were considered simultaneously. Only 5 variables were assessed by more than two studies. Inconsistent results were reported concerning four of the five remaining variables: maternal mental health and infant's health problems, gestational age and birth weight. Additionally, the way these variables were measured differed between studies, resulting in inconclusive data.

Table 3 Main findings of studies on the factors influencing the QoL of parents of preterm infants ($n = 12$)

Factors	Main findings
Mother	
Mental health	Higher EPDS scores: Lower mental maternal HRQOL ($P < 0.01$) [33, 34] Higher median BDI scores: Lower maternal QoL on physical, psychological, social and environmental domains ($P < 0.05$) [35] Higher Psychiatric Symptoms Index[a] scores: Poor overall maternal QoL ($P < 0.001$) [25]
Sleep	More severely disturbed sleep: Lower mental and physical maternal HRQOL ($P < 0.05$) [32–34] Higher daytime sleepiness: Lower physical and mental maternal HRQOL ($P < 0.01$) [32, 33]
Fatigue	Higher levels of fatigue: Lower physical and mental maternal HRQOL ($P < 0.01$) [33, 34]
Stress	Higher levels of stress: Lower mental maternal HRQOL ($P < 0.01$) [34]
Pregnancy complications	PT delivery (vs. Gestational hypertensive disorders or Gestational diabetes or. Uncomplicated pregnancy): Lower maternal QoL on physical domain ($P < 0.05$) [28]
Time postpartum	Week 3 (vs. Week 1): Higher maternal QoL on health and functioning domain ($P < 0.001$) [27]
Light exposure	≥ 12 h: Higher physical maternal HRQOL ($P < 0.01$) [32]
Religion	Evangelical: Higher maternal QoL on social domain ($P = 0.019$) [35]
Infant	
Health problems	Receiving HOT: Lower maternal QoL on vitality and mental health dimensions ($P < 0.05$) [31] Posthemorrhagic hydrocephalus: Lower maternal QoL on psychological ($P = 0.010$) and social domains ($P = 0.001$) [35] Bronchopulmonary dysplasia: Lower maternal QoL on physical domain ($P = 0.005$) [35] Higher score for neonatal acute physiology with perinatal extension: Lower maternal QoL on social domain ($P = 0.027$) [35] Cerebral palsy or hydrocephalus (vs. Neurologically normal): More stress related to the unwillingness of the paediatrician to agree that child had a problem or needed referral for therapy, and more difficulties related to the physical work to care for child [23]
Gestational age	24–33 weeks (vs. 34–37 weeks or 38–41 weeks): Lower maternal QoL on psychological/baby domain ($P < 0.001$) [27] Lower number of gestational weeks: Lower maternal QoL on physical domain ($P = 0.010$) [35]
Birth weight	VLBW (vs. Full-term): Higher overall caregiver's QoL ($P < 0.05$) [25]
Gender	Female: Higher maternal QoL on environmental domain ($P = 0.011$) [35]
Family	
Support system	The contact with other parents of preterm children had a positive impact on family QoL, while the lack of support provided by extended family had a negative impact on family QoL [23]
Memories of neonatal period	Religion or optimistic philosophy of life during neonatal period had a positive impact on family QoL, while the guilt considered as a problem in adjustment influenced negatively the family QoL [23]
Values and relationships to child	High appreciation of their child had a positive impact on family QoL, while considering child more "spoiled" or more protected by parents had a negative impact on family QoL [23]
Perceived impact on parents' lives	No much life changes influenced positively family QoL, while the difficulties related to the physical work to care for child had a negative impact on family QoL [23]
Plans for future children	No changes in the plans for future children had a positive impact on family QoL, while the deferral of the birth of later children or more care with later pregnancy had a negative impact on family QoL [23]
Family income	BRL 1500-2750[b]: Lower maternal QoL on environmental domain ($P = 0.001$) [35]
Marital union	Stable: Higher maternal QoL on social domain ($P = 0.004$) [35]
Health care	
Communication of medical information	Information and explanation of medical terms by the medical personnel had a positive impact on family QoL during neonatal period, and the absence of important medical information had a negative impact on family QoL [23]
Financial impact	Do not having problems with the neonatal hospital bill had a positive impact on family QoL, and problems with the costs of later medical care had a negative impact on family QoL [23]
Behaviour of medical personnel	Stress due to policy of transporting hospital had a negative impact on family QoL [23]
Hospitalization in NICU	During hospitalization (vs. 6 months after discharge or 12 months after discharge): Lower maternal QoL on physical domain ($P = 0.013$) [35]
Participation in a nutrition RCT	Enrolled (vs. Not enrolled): Higher parental QoL scores ($P = 0.02$) during infants hospitalization in NICU [30]

BDI Beck depression inventory, *BRL* Brazilian real, *EPDS* Edinburgh Postnatal Depression Scale, *HOT* home oxygen therapy, *HRQOL* health-related quality of life, *NICU* neonatal intensive care unit, *P* P value, *PT* preterm, *QoL* quality of life, *RCT* clinical randomized intervention trial, *VLBW* very low birth weight (< 1500 g)

[a] Depression, anxiety, anger and cognitive disturbance

[b] The equivalent to 406–745 €

Studies reported issues related with the family, the mother and health care as positively influencing parental perception of their own QoL. Having a stable marital union, maintaining contacts with other parents of preterm children, family religious belief or optimistic philosophy of life during the neonatal period, high appreciation of the child and having few changes in life and in plans for having future children, all had a positive impact on parents' QoL [23, 35]. Women experiencing a total light exposure of 12 or more hours per day [32], as well as women self-reporting evangelical religion [35], presented higher levels of perceived physical and social QoL than those who did not. Mothers' perception of QoL on health and functioning domains also improved over time during postpartum period [27]. Parents of preterm infants who participated in a Clinical Randomized Intervention Trial (vs. non-participating) reported significantly higher levels of QoL during hospitalization in NICU [30]. Having no problems with the neonatal hospital bill and receiving information and explanation of medical terms by healthcare professionals also had a positive impact on family QoL [23].

Low levels of QoL were mainly associated with mother-, family- and health care-related factors. Severely disturbed sleep, high levels of daytime sleepiness, fatigue and stress were associated with lower maternal QoL [32–34]. Family issues, such as family income of 1500–2750 Brazilian Reals (406 –745 €) [35], lack of support provided by extended family, feelings of guilt considered as a problem in adjustment, the difficult physical work to care for child, considering child more "spoiled" or more protected by parents and the birth of later children deferred or more care with later pregnancy [23], also had a negative impact on QoL. Hospitalization in NICU (vs. 6 or 12 months after discharge) [35], as well as absence of important medical information, stress due to policy of transporting hospital and problems with the financial costs of later medical care, constituted health care-related factors influencing negatively family QoL [23].

The circadian activity rhythms, parity, the number of miscarriages, night-time total sleep time and self-reported physical health, as well as maternal age, skin colour, educational level, occupation and working status, were not associated with maternal QoL [25, 26, 32, 33, 35]. Similarly, the length of infant's stay in NICU, the child's chronological age, the intrauterine growth restriction, the APGAR score at 5 min and the motor and language quotient were described as factors with no impact on parental QoL [25, 29, 31, 35]. The participation of the infant in a Clinical Randomized Intervention Trial during hospitalization had no impact on parental QoL at 3.5 years after delivery [30]. Finally, the QoL of mothers and fathers of preterm infants was not influenced by the mode of delivery, the number of prenatal care visits and the use of antenatal corticosteroids [28, 35] as well as by the family set up, the number of adults in home and the place of family residence [25, 26, 31].

Studies addressing infant factors related with the presence of health problems, gestational age, birth weight and gender revealed inconsistent results, as well as those assessing maternal mental health and pregnancy complications, which might be explained by differences on assessment tools and timing of data collection. Some studies revealed that higher mother's depression scores [33–35], caregivers' psychiatric symptoms [25] and pregnancy complications [28] were associated with lower QoL, while other reported no association between QoL and self-reported depression/anxiety or pregnancy-related diseases [35]. Having an infant born with 24–33 weeks of gestation (vs. 34–37 or 38–41 weeks), as well as with some health problems (receiving home oxygen therapy, posthemorrhagic hydrocephalus, bronchopulmonary dysplasia, high score for neonatal acute physiology with perinatal extension) was associated with worse maternal QoL [27, 31, 35]. Maternal QoL also tended to decline with the decrease of the number of gestational weeks at birth [35]. In contrast, other studies reported no difference between the QoL of mothers of preterm and full-term infants [26] and no association between infant's gestational age and parental QoL [31]. In addition, parenting an infant with cerebral palsy or hydrocephalus (vs. neurologically normal) revealed to be associated with more stress related to the unwillingness of the paediatrician to agree that the child had a problem or needed therapy and more difficulties related to the physical work to care for child, while had no impact on the problems with the neonatal hospital bill and the importance given to the support provided by extended family [23]. One study showed that the infant's ongoing medical problems were not associated with caregivers' QoL [25]. In addition, one study referred that having a very low birth weight infant (vs. full-term) was positively associated with parental QoL [25], while three other studies reported no association between infant's birth weight and maternal QoL [26, 31, 35]. Finally, one study found that having a female infant (vs. male) had a positive impact on maternal QoL [35], while another one revealed no association between infant's gender and maternal QoL [26].

Discussion

Current state of research and future direction

This scoping review suggested that the QoL of parents of preterm infants is mainly influenced by factors related with maternal characteristics, family issues and health care environment rather than aspects related with infants, in a framework where factors from individual fathers and structural levels were not addressed. Studies were based on specific sets of variables, for which the assessment varied among studies, with 40 out of 45 factors being analysed by only one or two studies.

Psychosocial characteristics of mothers, namely sleep disturbances, fatigue, stress and poorer mental health (in particular depression and psychiatric symptoms) proved the highest relevance as factors negatively influencing the QoL of mothers of preterm infants. Previous studies conducted during hospitalization in NICU consistently show that parents of preterm infants present high levels of depression, stress and anxiety [12, 14, 16, 36], and report a sense of uncertainty and powerlessness which conjointly impact negatively on parental sleep [37]. Thus, the findings of this scoping review emphasize the need for healthcare professionals to be aware of the impact of a preterm delivery on maternal mental and psychosocial health and sleep patterns during early years. This knowledge will help health professionals to identify groups at risk that should constitute a privileged target for early intervention, aiming to promote parental mental health and improve parents' sleep quality, especially during the first weeks' postpartum. According to our results, future research should explore the maximization of the hours of natural light exposure [32], and the coping strategies related with parents' religion [23, 35] as two possible strategies to increase parental QoL.

Five studies addressed family-related variables, but only two found any associations [23, 35]. They described the social support provided by partner, extended family and other parents of preterm children as a factor positively influencing parental mental health and QoL, reinforcing the importance of the support system also reported by parents of full-term infants [38–41]. At the same time, results coherently showed that parental QoL benefits from the fact that parents did not perceive a preterm delivery as a disruptive event for the family. In a context where a preterm delivery constitutes a risk factor for recurrence in subsequent pregnancies [42], parents tended to change their plans for reproductive trajectories when their first pregnancy ended with a preterm delivery [43], which may have a negative impact on family QoL. These findings call for the need to explore in depth the role of several coping strategies to handle adversity and to deal with a preterm delivery [44, 45] as a factor influencing positively parental and family QoL. Furthermore, there is room to explore the influence of other family-related factors on QoL of parents of preterm infants, including those which were addressed by only one study (e.g. family set up, number of adults at home and place of family residence).

Only four studies reported issues related with health care [23, 28, 30, 35]. The way medical information is communicated and the degree of parental concern with financial costs of medical care constituted two main factors related with health care environment influencing parental QoL. These results suggest that health professionals' acknowledgement of parental needs for information, financial support and assurance [46] when dealing with mothers and fathers of preterm infants is central to the development of integrated, sustainable and quality family-centred health care services. This review also highlights the importance of further exploring the impact of medical insurance and family income on parental QoL, in a context where caregivers/families of infants with physical and mental illness reported better QoL when having a public insurance [40, 41] and parents of preterm infants reported less emotional burden when they have financial compensation for time taken off from work [47].

Studies performed with parents of infants with chronic diseases also suggest that other variables than infant-related factors influence the QoL of mothers and fathers, in particular factors related with characteristics of mothers and fathers and health care environment [41, 48, 49]. However, some of these factors were not tested by most of the studies included in this scoping review, namely those associated with parents' self-efficacy and coping strategies [41, 50], maternal and paternal educational level [51] and employment status [52], family socioeconomic status [51], quality of the marital relation [40], parental health-related behaviours such as eating habits and exercise [49], use of community-based developmental resources (e.g. early intervention programs) [47] and regulation of parental leave [48]. The assessment of these variables in future research will contribute to a better understanding of QoL of parents of preterm infants.

Methodological features

There are some methodological limitations in the studies included in this scoping review that should be taken into account when analysing the results. A considerable heterogeneity across the studies was observed regarding the operationalization of QoL and the use of different units of analysis (mothers, parents, families and caregivers). Additionally, a small number of studies conducted in few countries and specific settings, with different periods of data collection, are available.

QoL, as defined by WHO [20], has been subjected to several interpretations. Only two studies presented a definition of QoL [25, 27], and the remaining used interchangeably concepts as health-related QoL, life satisfaction, parental functioning and well-being as proxies of QoL [28–30, 35]. This translates into the use of seven different quantitative instruments to measure the construct, which assessed specific domains and proxies of QoL. The presented findings can thus be biased by the different measures used to assess QoL [29, 32–34, 53].

In the last four decades, condition-specific QoL questionnaires have been developed for caregivers of infants with several medical conditions, aiming to assess the specific impact of each condition on caregivers' QoL [54]. However, no specific tool has been designed to assess QoL of parents of preterm infants. Although the failures of the available quantitative instruments were acknowledged in one study [26], no attempts were observed to explore whether there are dimensions or constructs specifically relevant for parents of preterm infants not addressed in the questionnaires. As pointed out in this scoping review, parents bring up some dimensions influencing their own QoL that differ from those covered by the quantitative instruments, namely the importance of the support system, the information needs and the medical, reproductive and social costs related with a preterm delivery. These findings call for the development of more mixed methods studies, which would lead to a wider understanding of the QoL of parents of preterm infants [55], assisting us to disentangle the mechanisms behind some contradictory findings, and to the identification of the issues that are missing from the available scales by involving different stakeholders (e.g. parents, health professionals and relevant community stakeholders) [53].

Finally, more detailed data about the QoL of fathers of preterm infants are required. It could serve as a basis for exploring if parenthood is more consistently linked to well-being among men than women [56]. Although fatherhood has been associated with greater life satisfaction, happiness, positive affect and less with depressive symptoms [57–59], literature suggested that fathers of preterm infants, similarly to mothers, experienced high rates of psychological distress after birth due to the simultaneously concern for the hospitalized infant, providing support to the mother, communicating with family and friends, caring for other infants and returning to work [12, 14, 60].

Conclusion

The aim of this scoping review was to synthesize the current body of knowledge on the factors influencing the QoL of mothers and fathers of preterm infants. Studies addressed mainly mother- and infant-related factors. The results suggested that parental QoL after a preterm delivery is mainly influenced by factors related with mother's characteristics, family issues and health care environment rather than infants' variables, in a context where factors regarding fathers' characteristics and structural levels were not addressed. There is a need for standardizing the operationalization of the QoL and developing a structured questionnaire adapted to the specific needs of mothers and fathers of preterm infants. Further research on parental QoL after a preterm delivery should include both mothers and fathers, invest in mixed methods approaches and be performed in different countries and settings for allowing integration and comparison of findings.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

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1.3. Family Integrated Neonatal Care

1.3.1. From inception to contemporary implementation

In the first half of the 20th century, the organization of paediatric wards routines was based on the assurance of practical needs of health professionals, with parental roles being confined to the delegation of care and decision-making around their children's health for the healthcare team (128). The clinical practice was then mainly centred on the control of infections, order, discipline and asceticism, neglecting the social and psychological needs of children and their parents, the latter often excluded from hospitals for fear of contagion (128). Reports of individuals hospitalised in British hospitals during childhood, between 1920 and 1970, confirm the absence of contact with their families during hospitalisation, in a context where health professionals did not manifest affection during clinical interactions (129). A patient-doctor relationship drawn upon an emotional distance characterizes a paternalistic model of care, according to which health professionals' authority lies exclusively on technical knowledge (130, 131).

A concern with the consequences of parental separation during the infant's hospitalisation period for both children and parents' health emerged after the Second World War (128, 129), mainly driven by the works of John Bowlby (132) and James Roberston (133). These studies had shown that young children separated from their mothers developed more frequently emotional, psychological and developmental problems (134, 135).

Thereafter, a shift on the attitudes and practices surrounding the presence of parents during their infant's hospitalisation period was observed (128, 134-137). Traditionally excluded from hospitals and paediatric services until the 50's, the family started to increasingly assume a central role in the health promotion and well-being of hospitalised children (73, 138). Civic movements as well as parental and professional associations, especially in the USA and the UK, joined efforts to have more child-friendly hospitals, advocating the extension of visiting hours and openness to host mothers during the night at the hospital (128, 136, 139). Several models of healthcare were developed for this purpose, such as the *parental participation* (140), the *care-by-parent* (141) and the *partnership-in-care* (142), all of them precursors of the *family centred care* (128) and *Family Integrated Care (FICare) models* (83). A pioneering example for the case of preterm and sick full-term infants was registered at Tallinn Children's Hospital in Estonia in 1979 (143), where a mother-baby unit was created for teaching mothers how to provide 24-hours care for infants with healthcare staff assistance in technical procedures, while promoting breastfeeding, minimal use of technology, and little contact between the baby and health professionals. This model of care resulted in a decrease on the number of infectious diseases, on the duration of intravenous infusions and antibiotic therapy, as well as on the improvement of infant's neurological development (143). Despite the criticism of some health professionals who saw parents as a negative factor in the care of hospitalised children (128, 138), those models succeeded in spreading the idea of family involvement during the hospitalisation period (144).

Meanwhile, a move towards patient centred care progressively challenged the predominant paternalistic model of healthcare delivery. This change represented a significant shift in terms of patients' participation in decision-making and care by conceiving high-quality healthcare as respectful of and responsive to individual patient preferences, needs and values (145-147). This transition was influenced by the psychoanalytical and psychosocial theories from Breuer and Freud (1955), by the person-centred therapy approach developed by Rogers (1961), as well as by the work developed by Balint (1964) around the importance of analysing the psychological and social context of patients and developing a single emotional relationship with them (148-150). Patients' experiences became relevant to healthcare systems and patients started to be seen as active agents in the management of their own health and health services (151), including the right for receiving comprehensive information, being treated with respect as well as being actively involved in the decision-making processes around their own medical treatments (152). In paediatric medical services, these principles are also applied to parents and families (153, 154).

Family centred care was initially conceived by parents and health professionals for children with special care needs, being formally defined for the first time in 1987 as medical care based on a respectful, collaborative and supportive relationship between families and health professionals (155). A focus on family experiences and needs and its involvement in the design and implementation of healthcare provision and decision-making processes sustained the emergence of family centred care as a governance model in paediatrics and, in particular, in neonatology, in the beginning of the 21st century (73, 93, 128, 156). This approach advocates the promotion of individualized and flexible care, suitable to the specific needs of each family, being mainly grounded on the provision of information and support, and on the establishment of effective communication between healthcare professionals and family members (136, 137, 157, 158). In summary, the family centred care model relies on the following principles (73, 124, 153, 159, 160):

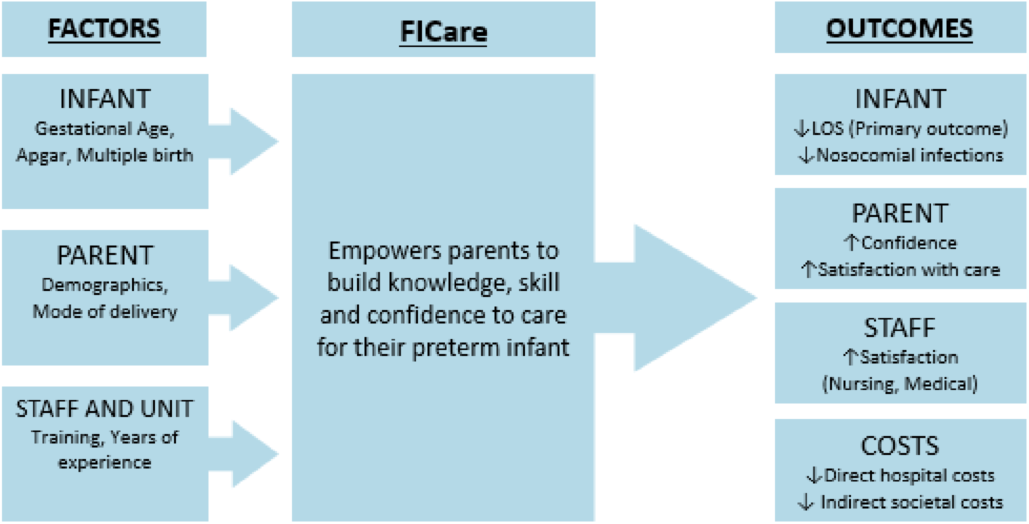
- Consider the family as a constant in the child's life;
- Respect the family racial, ethnic, cultural and socioeconomic diversity as well as its strengths, idiosyncrasies and coping strategies;

- Promote cooperation between parents and health professionals at all levels of care, including the bidirectional, complete and impartial sharing of information;
- Provide and/or ensure formal and informal social support;
- Adopt flexible health policies and practices tailored to each family's specific needs.

In 2001, the Committee on Quality of Health Care in America, from the Institute of Medicine, established the family centred care as a crucial indicator on the quality assessment of health services (161). Two years later, the American Academy of Pediatrics included this model as the standard of healthcare for all children, recognizing its innovativeness in the planning, provision and assessment of healthcare, thereafter based on a partnership between the family and healthcare providers that mutually benefits patients, families, healthcare staff and the health system (73, 159, 162). Arguments in favour the implementation of family centred care in NICUs were: reduction of infant's length of stay and number of readmissions (126, 163, 164); decline of morbidity associated with low birth weight (124, 165); decreased parenting stress levels and increased family satisfaction with care, improving assurance on healthcare staff and promoting confidence on parenting skills (126, 166); increased satisfaction levels among health professionals regarding work and performance (73, 124); increased hospital reputation, improving the capacity to hire and maintain NICUs professionals (72); and improvement of care services and health outcomes, potentially decreasing healthcare costs (73).

NICUs health professionals then reinforced the promotion of the kangaroo care or skin-to-skin care (124, 167), an early, prolonged and continuous skin-to-skin contact between the mother (or father) and the baby (168). This program of care became famous particularly because it does not need sophisticated equipment and it can be applied in several settings, including peripheral maternity units in very low-income countries, while potentially contributing to the humanization of neonatal care and cost savings (168, 169). However, kangaroo care still reproduced the idea that only NICUs professionals had the adequate technical skills to provide the majority of care for the infant (83).

Inspired by the evidence generated from experiences of family centred care and the Estonian human neonatal care model (143), a Canadian team of parents and healthcare professionals collaboratively developed the FICare model (Figure 3), aiming to treat the whole family (parents and their preterm infant) as a single unit of care (83). The FICare deepened the family centred care model by enabling parents to become integral members of the NICU team through support and empower them for being their infant's primary caregiver (71, 83). The four pillars of FICare model include: 1) *doctors and nurses education and support* in order to provide staff with the skills that enable them to educate, mentor and support parents in caring for their infant; 2) *a comprehensive parent education program* to provide parents with the skills and tools they need to confidently and safely care for their infant in the NICU and after discharge; 3) *adaptation of the NICUs policies, procedures and infrastructures* to families, enhancing their participation on infant's care; and 4) *psychosocial support* provided by families, peers, social workers and mental health professionals, to enable parents to overcome their fear and engage as a partner in care (83, 170-172).



Note: FICare – family integrated care; LOS – length of stay

Figure 3. Family Integrated Care Model (173)

According to the FICare model, infant, parent, and staff and unit factors may influence its implementation, with a focus on the analysis of maternal characteristics (173) while neglecting the extended family, the community and the country's setting and development status (83). It is expected that FICare positively impacts on outcomes related with infants (e.g. improve weight gain, increase the high-frequency of exclusive breastfeeding at discharge, decrease length of stay (LOS) and nosocomial infections), parents (e.g., decrease parenting stress and anxiety, increase their confidence and competence to care for preterm infants at home), staff (e.g., less conflicts with parents and greater job satisfaction), and healthcare costs (e.g., cost savings by reducing the use of resources, including duration of oxygen therapy and length of hospital stay as well as post-discharge support, outpatient clinic visits and re-hospitalisations) (71, 84, 173). Beyond child-centred and mental health parental outcomes (83), further research should include parental QoL (174) and assess the influence of both maternal and paternal characteristics, extended family/community and the macro-structural levels on the implementation of FICare inside and outside NICUs. Actually, a medium and long-term objective of FICare is maintaining parents as an integrated part of infant's care team after the NICU discharge. The mentors of this model, Karel O'Brien and Shoo K. Lee, argued that parents are able to deliver early interventions for their child at home with community help, by providing peer-to-peer support, community events for parents of preterm infants, online resources, mental health assessments and continuing parent education (175). In this sense, FICare aligns with the World Health Organization global strategy on people-centred and integrated health services (Figure 4) which advocates seeing individuals, families and communities as participants as well as beneficiaries of trusted health systems (176).

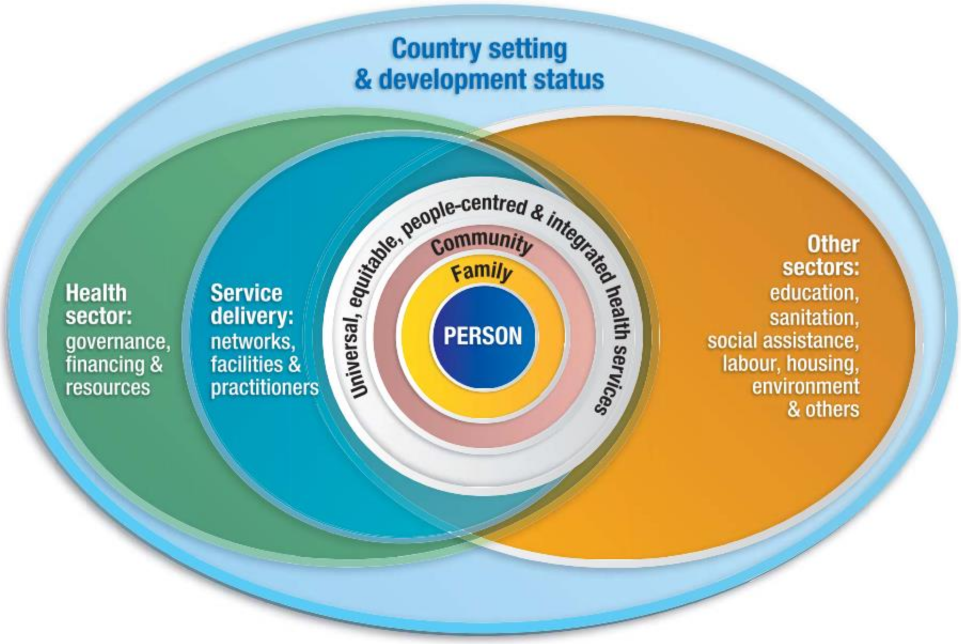


Figure 4. Conceptual framework for people-centred and integrated health services (176)

This renewed approach to healthcare provision and governance calls for shared information, shared engagement and shared accountability (127, 177), which may entails individual, organizational and political challenges. There is a need for some adjustments in the established roles, attitudes and knowledge that traditionally sustain the interactions between professionals, patients and families in healthcare systems; changes in the cultural, physical and operational characteristics of the NICUs; willingness of health professionals to be involved and improve their communication skills; and delivering care tailored to parental characteristics, preferences and choices (118, 175, 178). The literature discusses challenges for implementing FICare within NICUs' context and culture mainly based on healthcare professionals' views (175, 179), but the analysis of political and social issues is scarce with few studies acknowledging parents' point of view (175).

1.3.2. Parental involvement in FICare: Missing links

Few criticisms around the possible risks and limitations of family integrated neonatal care both for families and health services emerged in the literature. This discussion is particularly relevant when considering the dominant idea of an intensive parenthood in NICUs, according to which parenthood is emotionally involving, wholly child-centred and guided by specialists (180), concealing possible constraints associated with the legal and political context underlying the regulation of parenthood, and the complexity and diversity of parental experiences and needs (181, 182). The understanding of the opportunities and limitations associated with FICare from the perspective of mothers and fathers of infants hospitalised in a NICU is therefore fundamental for rethinking governance in neonatology (175, 183).

Health professionals tend to consider that FICare aims to delegate on families a greater responsibility for care and decision-making in relation to the child (184), while some parents feel uncomfortable and pressured to stay with and care for their infants, which they consider to be nurses' responsibility, reporting little or no support from them (138, 182, 183). Thus, it is important to analyse the practices and uses of FICare, exploring if they result from a process where parental and families’ expectations were taken into account. The low educational level of parents, the lack of support or the existence of financial constraints, as well as racial/ethnical differences and/or linguistic divergences between families and health professionals may also hinder the establishment of partnerships around the infant’s care (159). Moreover, the need for fulfilling work commitments to guarantee the family's financial assistance as well as the household composition and dynamics, such as single parenthood or the existence of dependent individuals at home demanding care, may constrain the presence and involvement of mothers and fathers in NICUs, in a context where parental absence often results in self-blame processes and professionals’ negative attitudes (138, 182).

Actually, the sustainability of FICare depends on social and political support, in particular regarding family-friendly and gender-equality policies. In contexts where the financial assistance during infant’s hospitalisation is insufficient, parents may be struggling with being present in the NICU, increasing health inequities. In non-Western countries, young families receive much less public support than in Organisation for Economic Co-operation and Development (OECD) countries, of which the Nordic countries have the most extensive and the USA and Australia the least extensive supports (e.g., available and affordable daycare, flexible work schedules, job leave security, cash benefits, and paid parental leave) (185, 186). Thus, further studies should explore societal factors influencing the implementation of family integrated neonatal care through the views of parents, especially during the return-to-work period, which is a relevant moment in countries where few attempts are being made to support parental leave (181, 187).

Moreover, FICare demands effective communication and negotiation skills from health professionals, but these subjects are mostly absent from their medical studies curriculum. Although the new principles of public management applied to health systems meet the patients' demands (188), they also aim to maximize a rational and efficient performance at the lowest cost (189) and push the professionals towards productivity objectives (190), weakening a truly commitment to FICare. Allied with the lack of communication and negotiation skills, the time, human and material resources constraints observed in NICUs highlights a deficiency of organizational and political support to FICare, fostering the demotivation of healthcare professionals (138, 159). The development of specific guidelines for health professionals would be helpful (191). However, major international guidelines in neonatology are only focused on some of the principals of FICare, such as family presence, parent education and participation in care, parent support, NICU environment, staff education and support (73, 118, 192-195), or on its applicability in specific moments, like breastfeeding (196, 197), kangaroo care (198, 199), palliative care (200-202), the preparation to discharge (193) and post-discharge support (118). Furthermore, the development of guidelines is more based on the perspectives of specialists than the experiences and viewpoints of mothers and fathers of infants hospitalised in NICUs (157). At a national level, the Portuguese NICUs guidelines include mainly parents' information needs, child care activities performed by parents and their responsibilities in decision-making regarding infant's health, while psychosocial consequences of parenting a child in NICUs, and the adequacy of their environmental characteristics to parental needs were less frequently touched upon (191). Parental and family characteristics are mentioned as risk factors for prematurity and perinatal diseases, but issues related to parental safety and comfort, and the confidence of parents in healthcare and social support are rarely mentioned. Thus, it is central to include the perspectives of both mothers and fathers into family centred-based practical guidelines for consistent and comprehensive collaboration between mothers, fathers and healthcare professionals in the improvement of providers' cultural sensitivity in counselling high-risk families.

2. Objectives

A very preterm delivery and the ensuing hospitalisation of the infant(s) in a NICU are considered disruptive, emotionally traumatic and stressful life events affecting parental QoL via multiple pathways (51, 114, 203, 204). Parenting a very preterm infant is related to an increased risk for developing parenting stress (75, 85, 205), depressive symptoms and anxiety shortly after delivery (76, 117), as well as poor family functioning and a high family burden several years after childbirth (206, 207). Most studies have excluded fathers and neglected the analysis of structural factors, offering a one-sided perspective that fails to consider the impact of prematurity on both parents and family or to assess dimensions related with shared governance for health, limiting the opportunities of co-producing family integrated healthcare systems (82, 97, 127, 157). Furthermore, there is a need for developing sensitive instruments adapted to the singular experience of parenting a very preterm infant, which may be improved when captured through mixed methods research (36, 40). Such an interdisciplinary approach enables researchers to disentangle the mechanisms behind some contradictory findings and to identify issues that are relevant for mothers, fathers and families.

Thus, assessing the QoL of the parents of very preterm infants and associated factors require a detailed analysis of their experiences, perspectives, needs and sources of stress not only during NICU hospitalisation, but also after discharge. Such in-depth parental perspectives are key, especially during the return-to-work period, which is a relevant moment in countries where few attempts are being made to support parental leave (181, 187), allowing the identification of parents at risk for immediate and extended physical and emotional burden and the reduction of social inequalities and stress in caring for a very preterm infant. This knowledge is essential to develop family integrated healthcare services and policies in relation to prematurity (83, 84) to the benefit of parents, children, their families (71, 72) as well as healthcare staff and health services (73).

Incorporating parents' perspectives on the organization of care and governance is especially relevant considering the observed mismatch between the perceptions of health professionals and parents on family needs in NICU (106, 111). These phenomena could lead to a gap between the care expected by parents and the care actually provided (112), as well as discrepancies between current guidelines and the families' actual needs (191). Mapping out parents' perspectives simultaneously related to shared care and governance is thus crucial for promoting quality healthcare systems and services centred on families and enabling their involvement in the co-production of health in neonatology (127).

Focusing on family integrated healthcare, this thesis aims to broaden the knowledge on parental QoL following a premature birth to sustain the development of policy and practice in neonatology by engaging with the following research question:

How do individual, familial, socioeconomic and political characteristics intertwine with the perceptions of mothers and fathers of very preterm infants on their QoL, during and after infant's hospitalisation in a NICU?

This mixed methods study will explore sources of stress, needs and QoL of mothers and fathers of very preterm infants, serving as a foundation to address the following specific objectives:

1. To identify sources of stress in mothers and fathers of very preterm infants hospitalised in NICU, and their association with socio-demographic, obstetric and infant's characteristics.
2. To validate the NICU Family Needs Inventory for the Portuguese population, and to propose a Short Form.
3. To explore the needs of mothers and fathers of very preterm infants hospitalised in NICU according to their socioeconomic position, obstetric history and infant's characteristics by integrating quantitative and qualitative data.
4. To explore both mothers' and fathers' perspectives about their own QoL 4 months after a very preterm delivery, by integrating quantitative and qualitative data.

3. Methods

3.1. Study design

This work is based on an observational and longitudinal mixed methods study (208, 209), which protocol has been previously described elsewhere (210). A multistage design was used, involving the collection of quantitative and qualitative data at two different time points: 1) during a NICU hospitalisation period, 15 to 22 days after childbirth, using individual quantitative questionnaires; 2) after discharge, approximately 4 months after childbirth, using qualitative semi-structured couples-based interviews and individual quantitative questionnaires (Figure 5). This design was chosen with the intention to merge quantitative and qualitative data to develop a more complete understanding of the impact of a very preterm childbirth on parental QoL (40).

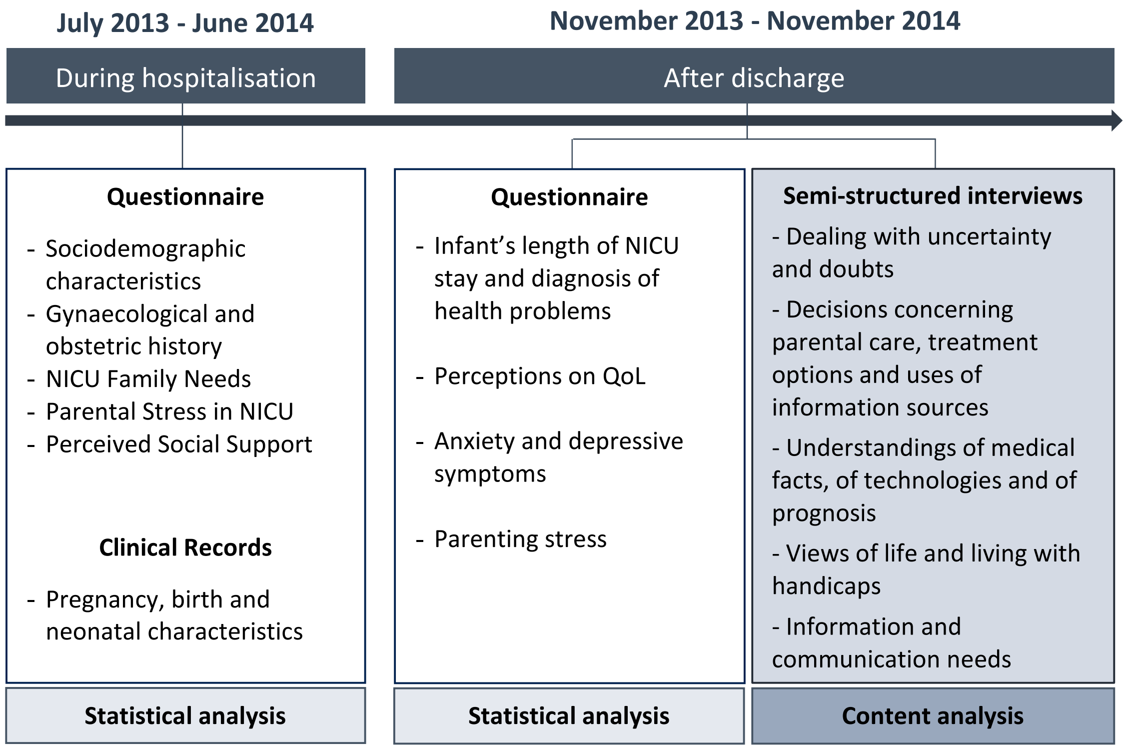


Figure 5. Study design

Evidence suggests that being alone or in the presence of one's partner shapes the reporting of experiences and emotions (211, 212) during data collection, with spousal presence leading to greater agreement on a variety of attitudinal and behavioural items (213). A primary issue considered was how to best approach the assessment of parental couples, individually or together (211). Quantitative questionnaires were administered to mothers and fathers separately in order to assess gender-specific lived experiences (214, 215) without the influence of one partner on the other, while respecting privacy and confidentiality as fundamental ethical principles (216). Semi-structured couples-based interviews were then conducted based on the rationale that including both parents would improve the understanding of processes by which marital relationship and gender roles are deployed to construct meanings of personal experiences on health (217), QoL and barriers and facilitators of FICare in NICUs (125). Moreover, recruiting participants at the couple-level increased the probability of fathers' participation (210, 211, 218).

3.2. Participants and data collection

3.2.1. Questionnaires during infants' hospitalisation

Between the 1st of July 2013 and the 30th of June 2014, mothers and fathers of very preterm infants (<32 weeks gestational weeks) hospitalised in all the 7 level III NICUs of the Northern Health Region of Portugal (Centro Materno-Infantil do Norte, Centro Hospitalar de S. João, Centro Hospitalar de Vila Nova de Gaia/Espinho, Unidade Local de Saúde de Matosinhos, Centro Hospitalar de Entre o Douro e Vouga, Hospital de Braga e Centro Hospitalar do Alto Ave) were systematically invited to participate in the study. Parents without serious illness that precluded NICU visitation (e.g., severe chronic conditions), who were present in the NICU during the hospitalisation period, who were able to speak and write Portuguese, and whose infants survived and were still hospitalised (not discharged or transferred to another hospital) at the time of the interview were considered eligible to participate in the study. Single families or individuals whose partners did not participate in the study were also considered eligible.

Parents were first approached by a NICU health professional (neonatologist or nurse), who was responsible for presenting the study and inviting participants to joint. All parents received an information sheet explaining the purposes and design of the study (see Appendix 1). When parents agreed to participate, the health professionals scheduled the most convenient date to administer the questionnaires. On the scheduled day, one member of the research team met participants at the NICU, responded to all their questions and doubts about the study, and accompanied them to a private room, where they read and signed the informed consent, and data collection was initiated.

Among the 126 eligible families, 122 families accepted to participate (participation rate = 96.8%), being included 120 mothers and 91 fathers (Figure 6): two mothers were absent because of medical complications, and 31 fathers were absent due to work commitments or emigration.

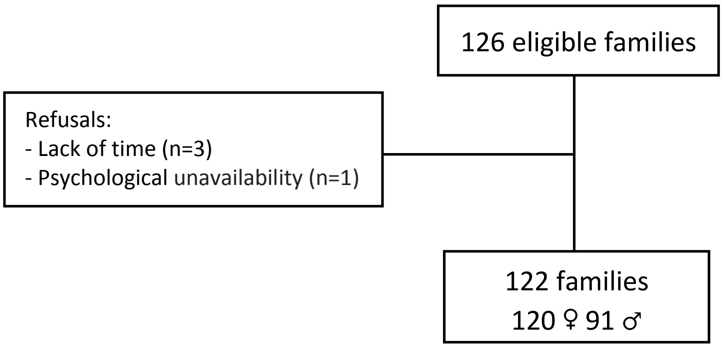


Figure 6. Flowchart for sample recruitment

Trained interviewers were responsible for conducting face-to-face interviews, using structured questionnaires developed by the research team (Appendix 2), to mothers and fathers separately but within the same timeframe. Parents were questioned 15 to 22 days after childbirth. This timing is based on results from a pilot study we conducted, which assessed the effect of data collection period on self-reported needs and stress among parents in NICU. A draft of the research protocol proposed the second week of infant's hospitalization as the period for assessing parental needs and stress through surveys; however, data obtained during ethnographic observation, conducted before the administration of the questionnaires, indicated the possibility to post pone the assessment to 15 to 22 days after child's admission for minimizing the burden to participants. A comparison of those interviewed at 8 to 14 days (10 mothers and 9 fathers) with those interviewed at 15 to 22 days after childbirth (12 mothers and 8 fathers) revealed that parents questioned within the 15-22 day bracket were less likely to refuse participation and they had a deeper awareness of parents-centred needs (support and comfort) and stress caused by changes in parental roles. These results supported the idea that the third week after childbirth constituted the appropriate moment for data assessment for research purposes (219).

The questionnaire included two parts. Firstly, five interviewers[1] collected data on sociodemographic characteristics (age, educational level, marital status, occupation, household monthly income and subjective social class) as well as on number of previous children. Afterwards, participants filled in the self-administered questionnaires regarding perceived social support, parental needs in the NICU, and parental stress. In addition, interviewers reviewed the clinical records of NICU hospitalisation to retrieve data on pregnancy complications (including infectious, placental, haemorrhagic and cardiovascular complications), mode of delivery, multiple pregnancy and neonatal characteristics of infants (birth weight and gestational age).

Parental occupations were classified by major professional groups, according to the Portuguese Classification of Occupations (PCO) 2010 (220) and then grouped in three categories: upper-white-collar, lower-white-collar and blue-collar occupations. The upper-white-collar category comprised individuals classified in the upper three major groups of the PCO 2010: executive civil servants, industrial directors and executives; professionals and scientists and middle management and technicians. The lower-white-collar category comprised individuals classified in the fourth and fifth major group of the PCO 2010: administrative and related workers and service and sales workers. The blue-collar category comprised individuals classified in the sixth to ninth major groups of the PCO 2010, including farmers and skilled agricultural, fisheries

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[1] The PhD candidate conducted over half of the structured interviews (n=107).

workers, skilled workers, craftsmen and similar, machine operators and assembly workers and unskilled workers. Students (n=2), housewives (n=4) and armed forces occupations (n=3) were excluded from this classification. Unemployed (n=36) or retired participants (n=1) were classified considering their previous main occupation, when mentioned.

Participants were georeferenced according to home address, using the ArcGIS Online World Geocoding Service and Google Maps. This procedure allowed matching each participant to the following socioeconomic contextual variables: urbanity level, neighbourhood socioeconomic deprivation and distance in minutes from residence to NICU. Urbanity level was determined according to the classification of urban areas, published by the Statistics Portugal in 2014. This classification groups the Portuguese parishes ('freguesias') into three classes: predominantly urban, moderately urban and predominantly rural areas (221). The European Deprivation Index (EDI) was used to classify the neighbourhoods according to their level of socioeconomic deprivation. EDI is a transnational multivariate index developed for five European countries (France, England, Italy, Spain, and Portugal), constructed using both individual and area level census data (222). The index was then categorized into tertiles, from tertile 1 (T1 - least deprived) to tertile 3 (T3 - most deprived). The shortest road distance from the participant's residence to NICU was calculated using ArcGIS version 10.4.1 and the Network analysist extension (223). The street network, required to calculate road distances, was provided by courtesy from Environmental Systems Research Institute (ESRI).

The characteristics of participants, stratified by gender, are presented in Table 1. When compared to fathers, mothers presented significantly higher levels of education and were more likely to have upper- and lower-white-collar occupations.

The Portuguese version of the *Multidimensional Scale of Perceived Social Support* (224) was used to measure the perceived adequacy of social support received from a significant other, family and friends, through 12 items on a 7-point Likert scale ranging from 1 (completely disagree) to 7 (completely agree). Each subscale has four items and higher values represents better perception of social support. The Portuguese version of the scale has shown good internal consistency (α=.94 among women and α=.93 among men for the general population).

Table 1. Characteristics of the participants, stratified by gender.

	Mothers (n=120)	Fathers (n=91)	*P* value
Age, years			
<35	85 (70.8)	63 (69.2)	
≥35	35 (29.2)	28 (30.7)	.801
Educational level, years			
≤12	68 (56.7)	64 (70.3)	
>12	52 (43.3)	27 (29.7)	**.042**
Marital status			
Single/Divorced/Widower	16 (13.3)	11 (12.1)	
Married/Living with a partner	104 (86.7)	80 (87.9)	.789
Occupation[a]			
Upper white collar	44 (40.4)	39 (44.8)	
Lower white collar	41 (37.6)	16 (18.4)	
Blue Collar	24 (22.0)	32 (36.8)	**.007**
Household monthly income, €			
≤1000	45 (38.5)	27 (29.7)	
>1000	72 (61.5)	64 (70.3)	.186
Subjective social class			
Low/Medium-low	87 (74.4)	77 (84.6)	
Medium-high/High	30 (25.6)	14 (15.4)	.072
Distance from home to NICU, minutes			
<15	58 (50.0)	48 (53.9)	
≥15	58 (50.0)	41 (46.1)	.577
Neighbourhood socioeconomic deprivation			
T1 (Least deprived)	48 (41.4)	37 (41.6)	
T2	34 (29.3)	25 (28.1)	
T3 (Most deprived)	34 (29.3)	27 (30.3)	.978
Urbanity level			
Predominantly rural/Moderately urban	17 (14.7)	9 (10.1)	
Predominantly urban	99 (85.3)	80 (89.9)	.333
Previous children			
No	85 (70.8)	67 (73.6)	
Yes	35 (29.2)	24 (26.4)	.654
Multiple pregnancy			
No	92 (76.7)	69 (75.8)	
Yes	28 (23.3)	22 (24.2)	.887
Pregnancy complications[b]			
No	70 (58.3)	51 (56.0)	
Yes	50 (41.7)	40 (44.0)	.739
Mode of delivery			
Vaginal/Instrumental	38 (31.7)	27 (29.7)	
Caesarean section	82 (68.3)	64 (70.3)	.756
Extremely low birth weight delivery[c]			
No	83 (69.2)	65 (71.4)	
Yes	37 (30.8)	26 (28.6)	.772
Extremely preterm delivery[d]			
No	95 (79.2)	71 (78.0)	
Yes	25 (20.8)	20 (22.0)	.841

Notes: IQR – Interquartile range; Data are n (percentage); In each variable, the total may not add 120 mothers or 91 fathers due to missing values; The proportions may not add 100 due to rounding.

[a]Students, housewives and armed forces occupations were excluded; [b]Infectious, placental, haemorrhagic and cardiovascular complications; [c]<1000g; [d]<28 gestational weeks.

The *NICU Family Needs Inventory* (110), a self-report scale consisting of 56 items, was applied to measure the importance attributed to family needs. Each item ranges from 1 to 4 (1 - not important, 2 - slightly important, 3 - important and 4 - very important), being grouped into 5 subscales: “Assurance”, “Proximity”, “Information”, “Support” and “Comfort”. At the end of the inventory, parents can describe other needs not addressed in the inventory and score them using the same scale in an open-ended question. The cultural adaptation and validation of the Portuguese version of this inventory is one of the specific objectives of this thesis.

The Portuguese version of the *Parental Stressor Scale: Neonatal Intensive Care Unit* (225), with 26 items, was used to measure parental perception of sources of stress arising from the environment of the NICU. Each item ranges from 1 (not at all stressful) to 5 (extremely stressful), being grouped into 3 dimensions: “Sights and Sounds” (6 items), “Baby Looks and Behaves” (13 items) and “Change in Parental Role” (7 items). At the end of the questionnaire, there is a question about “Overall stress”. The score of each dimension of the stress scale is calculated as the mean of the group of the respective individual items. It ranges from 1 to 5, with higher values indicating higher levels of parental stress. The Portuguese version of the scale presented good internal consistency (α=.80 for the subscale “Sights and Sounds” and α>.89 for the subscales “Baby Looks and Behaves” and “Change in Parental Role”).

3.2.2 Questionnaires after infants’ discharge

Approximately 4 months after childbirth, between the 1st of November 2013 and the 30th of November 2014, families who previously accepted to participate in the second phase of the study (n=117) were contacted by telephone or e-mail, according to their preference, to confirm their availability to receive the questionnaires at home. This timeframe was chosen because it is the common return-to-work period in Portugal, in particular for mothers, constituting a relevant moment in countries where few attempts are being made to support parental leave (181, 187). Parents whose infants were still hospitalised (n=1) or died (n=3) were not eligible to integrate this phase of the study. Self-administered questionnaires for individual completion, informed consent forms, with prepaid return envelops, were sent by mail. Parents who accepted to participate and did not return the questionnaire within approximately one month were reminded to do so, by telephone or e-mail, up to three times. Among the 113 eligible families, 67 families completed and returned the questionnaires (participation rate = 59.3%), corresponding to 67 mothers and 64 fathers (Median months after childbirth (P25-P75): 4.3 (4.0-4.6)). There were nine fathers participating in this phase of the study that were not assessed during infants' hospitalisation period.

Data on infant's length of NICU stay and diagnosis of health problems (inguinal and umbilical hernias, metabolic disease, ovarian cysts, bronchial dysplasia, autoimmune disease, cardiac disease, congenital malformation) were collected by self-report.

Symptoms of anxiety and depression as well as parenting stress and perceived QoL were assessed through scales validated in Portuguese samples. The *Beck Anxiety Inventory* (226) and the *Beck Depression Inventory-II* (227) consist of 21 items each, on a 4-points Likert scale, which are statements describing anxiety and depressive symptoms respectively, that respondents evaluate with reference to their own state. Higher values indicate higher levels of anxiety and depressive symptoms (ranging from 0-63). The Portuguese version of the *Beck Anxiety Inventory* has shown reasonably good internal consistency (α=.79) and of the *Beck Depression Inventory-II* presented good internal consistency (α=.91).

The *Parenting Stress Index* (228) is a 104-item inventory on a 5-point Likert scale (ranging from 1 - completely agree to 5 - completely disagree) plus a Life Stress scale (composed of yes/no statements) designed to evaluate the magnitude of stress in the parent-child system. It is composed by two domains: "Child domain" (6 subscales) evaluating sources of stress as gathered from the parent's report of child characteristics, and "Parent domain" (7 subscales) measuring sources of stress related to parent characteristics. The total stress score is the sum of the scores in those two domains, with higher values indicating higher levels of parenting stress (range for the total scale: 104 to 517). The life stress scale is composed of 24 items covering family contextual issues such as parental separation, loss of income and work-related problems, with higher values indicating more stress in life (ranging from 0 to 114)[2].

The *World Health Organization Quality of Life – BREF Inventory* (229) consists of 26 items on a 5-points Likert scale, assessing individuals' perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns. It is organized into a facet of overall QoL (general perception of QoL and health) and 4 domains: "Physical", "Psychological", "Social relationships" and "Environment". Higher values represent better QoL (range: 0-100). The Portuguese version of the inventory has shown good internal consistency (α=.92).

There were no significant differences between parents who participated and those who did not in the 4 months questionnaire regarding: socioeconomic factors; (gender, age, educational level, occupation, household monthly income, subjective social class, neighbourhood

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[2] This instrument is currently being validated for the Portuguese population by CEGOC©.

socioeconomic deprivation and urbanity level); obstetric characteristics (previous children, multiple pregnancy, pregnancy complications and mode of delivery), or the characteristics of the infants at birth (gestational age and birth weight). However, parents who participated in the 4 months evaluation were more likely to be married or living with a partner than those who did not (Table 2).

Table 2. Comparison between socioeconomic, obstetric and infants' characteristics of the participants and the non-participants at 4 months' evaluation.

	Participants (n=131)	Non-participants[a] (n=82)	*P* value
Gender			
Female	67 (51.2)	49 (59.8)	
Male	64 (48.9)	33 (40.2)	.219
Age, years			
<35	84 (67.2)	59 (72.0)	
≥35	41 (32.8)	23 (28.0)	.469
Educational level, years			
≤12	75 (59.5)	57 (69.5)	
>12	51 (40.5)	25 (30.5)	.144
Marital status			
Single/Divorced/Widower	10 (7.9)	14 (17.1)	
Married/Living with a partner	116 (92.1)	68 (82.9)	**.044**
Occupation[b]			
Upper white collar	53 (46.1)	28 (37.3)	
Lower white collar	33 (28.7)	24 (32.0)	
Blue Collar	29 (25.2)	23 (30.7)	.479
Household monthly income, €			
≤1000	43 (34.4)	28 (35.4)	
>1000	82 (65.6)	51 (64.6)	.879
Subjective social class			
Low/Medium-low	95 (76.6)	66 (82.5)	
Medium-high/High	29 (23.4)	14 (17.5)	.314
Neighbourhood socioeconomic deprivation			
T1 (Least deprived)	57 (43.5)	31 (39.2)	
T2	40 (30.5)	22 (27.9)	
T3 (Most deprived)	34 (26.0)	26 (32.9)	.557
Urbanity level			
Predominantly rural/Moderately urban	6 (4.6)	2 (2.5)	
Predominantly urban	125 (95.4)	77 (97.5)	.453
Previous children			
No	90 (73.8)	59 (72.0)	
Yes	32 (26.2)	23 (28.0)	.774
Multiple pregnancy			
No	104 (79.4)	58 (70.7)	
Yes	27 (20.6)	24 (29.3)	.150
Pregnancy complications[c]			
No	74 (56.5)	49 (59.8)	
Yes	57 (43.5)	33 (40.2)	.639
Mode of delivery			
Vaginal/Instrumental	35 (26.7)	28 (34.2)	
Caesarean section	96 (73.3)	54 (65.9)	.248
Extremely low birth weight delivery[d]			
No	92 (70.2)	59 (72.0)	
Yes	39 (29.8)	23 (28.0)	.788
Extremely preterm delivery[e]			
No	106 (80.9)	66 (80.5)	
Yes	25 (19.1)	16 (19.5)	.939

Notes: IQR – Interquartile range; Data are n (percentage); In each variable, the total may not add 131 participants or 82 non-participants due to missing values; The proportions may not add 100 due to rounding.

[a]Non-eligible parents are not included in the analysis (n=4); [b]Students, housewives and armed forces occupations were excluded; [c]Infectious, placental, haemorrhagic and cardiovascular complications; [d]<1000g; [e]<28 gestational weeks.

3.2.3 Semi-structured couple-level interviews after infants' discharge

Between November 2013 and April 2014, parents who had previously accepted to be contacted 4 months after childbirth were systematically invited to participate in a couple-based interview. Participants were purposively sampled to include parents of extremely low birth weight infants (<1000g) and parents of non-extremely low birth weight infants (≥1000g) (42), in accordance with the distribution of the quantitative sample. This option was based on evidence regarding the importance of infant's birth weight to parents as it is frequently used as a proxy of the degree of infant's vulnerability (206, 230), as well as on the results obtained during ethnography and the NICU-based survey regarding the influence of infant's birth weight on parents' experiences and QoL (231). Additionally, a heterogeneity sampling was used for maximum variation of views and experiences, until reaching the thematic saturation. Thus, recruitment continued until no new themes emerged from the interview data (232).

Among 49 couples invited, 23 refused to be interviewed by lack of interest in the study (n=12), unwillingness of one member of the couple to participate (n=5), infant's illness (n=3), lack of time (n=2) and psychological unavailability (n=1). Thus, semi-structured qualitative interviews were conducted with a subsample of 26 parental couples with a joint mode of interviewing. The characteristics of the interviewees are summarized in Table 3.

Each member of the couple signed an individual informed consent form, with a specific agreement for audio recording. Interviews were conducted by the same female interviewer (PhD candidate) at parents' home (n=19), at the university department responsible for the study (n=6) and in a private hospital room (n=1). Interview duration ranged from 20 to 72 minutes, with an average of 39 minutes. All interviews were taped, transcribed verbatim and accuracy has been checked.

The interview schedule was developed by the research team according to the objectives of the study and informed by previous literature in the area as well as by data collected during ethnography in one NICU (210). The topic guide covered the following issues: how parents of very preterm infants deal with uncertainty and doubts and how they made their decisions concerning parental care (namely in the fields of physical contact and breastfeeding), treatment options and uses of information sources; their views of the consent processes; their understandings of medical facts, of technologies applied to perinatal care and of prognosis; their views of life and living with handicaps; information and communication needs of parents; and their wider awareness of social and ethical issues in this area.

Table 3. Characteristics of the interviewees.

Interview[a]	Age (years)		Educational level (years)		Multiple pregnancy	Extremely low birth weight delivery[b]	Previous children	
	Mother	Father	Mother	Father			Mother	Father
I1	27	25	≤12	≤12	No	No	No	No
I2	28	30	>12	≤12	No	Yes	No	No
I3	31	33	>12	>12	No	Yes	No	No
I4	42	NR	≤12	NR	No	No	Yes	NR
I5	33	36	>12	>12	No	No	No	No
I6	27	NR	≤12	NR	No	No	No	NR
I7	36	34	≤12	≤12	Yes	No	No	Yes
I8	37	26	≤12	≤12	No	No	No	No
I9	35	NR	≤12	NR	No	No	No	NR
I10	35	31	>12	>12	Yes	No	No	No
I11	39	35	>12	>12	No	No	No	No
I12	33	31	>12	>12	No	No	No	No
I13	38	36	≤12	≤12	No	Yes	No	No
I14	40	42	≤12	≤12	No	No	Yes	Yes
I15	25	29	≤12	≤12	No	No	No	No
I16	26	28	≤12	≤12	No	No	No	Yes
I17	26	32	≤12	≤12	No	No	No	No
I18	30	31	≤12	≤12	Yes	No	No	No
I19	33	32	>12	≤12	No	Yes	No	No
I20	24	24	≤12	≤12	No	Yes	No	No
I21	33	35	≤12	>12	No	Yes	Yes	Yes
I22	35	27	>12	>12	No	Yes	No	No
I23	24	29	≤12	≤12	No	Yes	No	No
I24	28	33	>12	>12	No	No	No	No
I25	31	35	>12	>12	Yes	Yes	No	No
I26	33	38	>12	>12	Yes	No	No	No

Notes: NR - not reported.

[a] Participants are described in the table following the order of interview - the alphanumeric code assigned to each couple corresponds to the number of interview order; [b] <1000g.

3.3. Data analysis

For an in-depth analysis of the experience of parenting a very preterm infant inside and outside a NICU, two different mixed methods designs were used according to the objectives of each study: a sequential exploratory design and convergent single-phase design. The comprehensive mapping out of both mothers' and fathers' needs simultaneously related to shared care and governance in NICUs benefited from a sequential explanatory design, whereby the quantitative data were first analysed followed by an interpretation of qualitative data. The rationale for this approach was that quantitative results and their subsequent analysis provided a general understanding of the most valued needs and gender specific differences, and qualitative data refine, explain and expand those statistical results by exploring more deeply parental views, adding some needs not assessed in quantitative instrument (208, 233). A convergent single-phase design, where the quantitative and qualitative data were collected during the same timeframe and with equal weight, was chosen to understand the QoL of parents of very preterm infants during the return-to-work period, mainly due to the absence of an instrument specifically designed for this population. Aiming to merge quantitative and qualitative data into one overall interpretation, in which quantitative results were validated or expanded with qualitative data (40, 208), this design allows a more complete comprehension of factors influencing mothers' and fathers' QoL after a very preterm childbirth.

Quantitative data

Statistical analysis was performed using the following software: Stata® version 11.0 (College Station, TX, USA, 2009), R® Statistical Programming Language version 3.2.2 and MPlus version 5.2.

Data was described as counts and proportions for categorical variables, means and standard deviations for normally distributed continuous variables, and median and interquartile range for non-normally distributed continuous variables.

According to the specific objectives of each paper, different analytic approaches were considered. The chosen analytic approaches, as well as how missing data was dealt with, are described in detail in the methods section of the papers. The Chi-square test or Fisher's exact test were used to assess the independent association between the categorical variables (sociodemographic, obstetric and infants' characteristics) and the outcomes. For continuous variables (social support, parental stress in NICU, NICU family needs, depressive symptoms, anxiety symptoms, parenting stress and QoL) mean or median differences were compared using the Independent Samples t-test or the Mann-Whitney test, according to data distribution.

The associations between explanatory variables and the outcomes were estimated by crude and adjusted mean differences (β) ant the corresponding 95% CI using linear regression models, stratified by gender. Statistical significance was set at a value of $P < .05$.

Qualitative data

Semi-structured qualitative interviews were analysed using the software NVivo11 (QSR International, USA, 2015). A triangulation strategy was used to guarantee the rigour and quality of research – an independent analysis was conducted by two researchers to identify, sentence by sentence, parental needs experienced in the NICU as well as parental perspectives about the factors influencing (positively and negatively) their QoL after a very preterm childbirth. Thematic content analysis was performed according to the protocol established by Braun and Clarke (234). Firstly, quotations with similar meanings were synthesized into categories, both inductively and deductively (according to the objectives of each paper). Secondly, the categories were grouped into analytical themes. Internal reliability and reflection were maximized comparing coding between multiple researchers, as well as re-examining qualitative data when disagreements with quantitative results were found (235).

3.4. Ethics

In data collection, storage, analysis and dissemination, procedures were developed in order to guarantee data protection and confidentiality. The study protocol was approved by the National Data Protection Commission and the Ethics Committee from all the 7 hospitals where the study was conducted.

All participants formalized their collaboration through a written informed consent form according to the World Medical Association's Declaration of Helsinki. Each participant provided prior consent to obtain personal contacts, collect information from clinical records and interview recording.

All materials were anonymous and confidential. Each NICU and each participant was identified with a numerical code, in the questionnaires and in the databases. Data protection was guaranteed in accordance with the usual rules of confidentiality and only the research team has access to the data. Personal data, informed consent forms, questionnaires, interview tapes and transcripts were coded and kept separately from one another in locked file cabinets. All audio files were destroyed at the end of the study and interview transcripts will be archived by ISPUP for 5 years. Once archived, transcripts were subject to strict protection and were not available, unedited, to any second party.

The interviewers were trained using a structured protocol addressing all the questionnaires' queries and periodic supervision of their work were undertaken by senior researchers. A multidisciplinary team, with experience in national and international projects, was responsible for the staff training and the development of the questionnaire and the interview topic guide. Transcription of the interviews was done by a professional and reliable service with a strict confidentiality policy in operation. Identifiable information was inevitably captured on the audio recordings, however only the research team and transcription service have access to these files and all the names mentioned were substituted by alias in the transcription.

4. Results

4.1. PAPER II: Parenting very preterm infants and stress in Neonatal Intensive Care Units

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Parenting very preterm infants and stress in Neonatal Intensive Care Units

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ABSTRACT

Background: Assessing parental stress during infants' hospitalization in Neonatal Intensive Care Units (NICU) is essential to identify parents at risk for immediate and extended physical and emotional burden.
Aims: To identify sources of stress in mothers and fathers of very preterm infants hospitalized in NICU, and their association with sociodemographic, obstetric and infants' characteristics.
Study design: Observational and cross-sectional study conducted between July 2013 and June 2014.
Subjects: Parents of very preterm infants hospitalized in all level III NICU in the Northern Health Region of Portugal were consecutively and systematically invited to participate in this study, being included 120 mothers and 91 fathers (participation rate: 96.8%).
Outcome measures: The Portuguese version of the Parental Stressor Scale: Neonatal Intensive Care Unit was used.
Results: The overall experience of hospitalization was classified as more stressful than the median for the subscales. "Change in parental role" was classified as the most stressful subscale by mothers (Median (P25–P75): 4.1(3.2–4.7)) and fathers (Median (P25–P75): 3.2(2.4–4.0)). Mothers scored significantly higher in all subscales. For mothers, multiple pregnancy was associated with lower levels of stress regarding "change in parental role" ($\beta = -0.597$; 95% CI $= -1.020$ to -0.174) and "overall stress" ($\beta = -0.603$; 95% CI $= -1.052$ to -0.153). Being ≥30 years old was found to be a significant predictor for decreased fathers' stress.
Conclusions: This study raises awareness for the need to develop sensitive instruments that take notice of gender, social support and family-centered care. The implementation of interventions focused on reducing parental stress is crucial to diminish disparities in family health.

1. Introduction

The delivery of a very preterm infant, occurring at <32 gestational weeks [1], and his/her subsequent hospitalization in a Neonatal Intensive Care Unit (NICU), is often described as an emotional rollercoaster [2] and a stressful and disruptive life event for mothers and fathers [3]. Parenting a preterm infant' implies a continuous redefinition and adaptation of expected parental roles [4,5], while dealing with the loss of hope to give birth to a healthy neonate as well as of the "phantasy self-as-mother", an idealized state where no mistakes are ever made [6].

Throughout the hospitalization of very preterm infants in NICU, parents encounter multiple stressors that may interfere with the parent-infant relationship. First, the infant's medical condition and immaturity and his/her appearance, abnormal breathing and lower responsiveness to social interactions [4,7,8]. Second, the impediments to the development of interaction skills by both parents and the infant (e.g. the limited availability of the infant, parents' inability to focus on the infant's cues and to recognize his/her behaviors) that preclude changes in parental roles [9]. Third, the concern that the healthcare team may misunderstand the child's needs and the parents' feeling of lack of information on the diagnosis or treatment [8]. Fourth, the stressors related with the transition process to parenthood [2,10], alongside feelings of self-blame and guilt for putting the child through pain [11], which is

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particularly experienced by mothers [6,9]. Finally, the complexity of the NICU environment, with unknown specific smells and lights, noisy life support and monitoring equipment [4,7], lack of privacy [12] and the constant presence of healthcare professionals [13].

Several studies show that parental stress in NICU is influenced by a range of sociodemographic, obstetric and infant characteristics. These include parental age, ethnicity, socioeconomic status [14] and exposure to other stressful life events [15], as well as pregnancy planning and previous pregnancy loss, maternal trait anxiety and mental health history, severity of infant illness [2,10,16] and previous experience of infant's hospitalization in NICU [17]. However, the results of these studies are inconsistent. Moreover, most studies exclude fathers, offering a one-sided perspective that fails to approach the impact of NICU hospitalization on the parents and in the family. Furthermore, there are very few studies focusing on sources of stress among parents of very preterm infants and those that exist were mainly conducted in the United States of America [16].

Previous research also suggests that parents' experiences in NICU are associated with posttraumatic stress disorder beyond the period of hospitalization [2,13], with a very preterm birth influencing the family environment several years later. Studies report poorer family functioning and higher family burden 2 and 7 years after birth among preterm families when compared to families of term born infants [19]. Stressful family environments, stress experienced among couples and the potential long-term consequences of stress on parenting and child health over time may be exacerbated by the socioeconomic milieu [20]. Considering the association between social adversity across the life course and the development of non-communicable diseases [18,21], experiencing a very preterm delivery can have a longer impact on parents' health and well-being [18].

The assessment of sources of parental stress during infants' hospitalization in NICU and its associated factors is essential to identify parents at risk for immediate and extended physical and emotional burden. Knowledge about such stress sources and factors may help healthcare professionals to develop and implement measures and interventions aiming to provide benchmarks for quality improvement in NICU [13] and to promote family-centered care [22]. This study aims to help achieve these goals by identifying sources of stress in mothers and fathers of very preterm infants hospitalized in NICU, and their association with sociodemographic, obstetric and infant's characteristics.

2. Methods

This observational and cross-sectional study is based on a cohort of mothers and fathers of very preterm infants, which protocol has been previously described elsewhere [23]. The study was approved by the Portuguese Data Protection Authority and the Ethics Committees of all hospitals where the study was performed and written informed consent was obtained from all participants.

Briefly, all mothers and fathers of very preterm infants born between 1st July 2013 and 30th June 2014 and hospitalized in all level III NICU located in Northern Health Region of Portugal, 7 in total, were consecutively and systematically invited to participate in the study. Parents were approached during the hospital stay by a NICU health professional (neonatologist or nurse), who was responsible for the study presentation and invitation. A total of 201 very preterm infants were born, corresponding to 165 families (130 single pregnancies, 34 twin pregnancies and 1 triplet). After excluding families whose infants were not hospitalized in NICU at the time of the interview, due to discharge, transfer to another hospital or dead (n = 27), families with serious illness that precluded NICU visitation (e.g., severe chronic conditions) (n = 4), families who were not present in NICU during the baby's hospitalization period (n = 6), and those who did not read Portuguese (n = 2), 126 families were eligible to integrate the study. Among these, 122 (96.8%) accepted to participate. The study included 120 mothers and 91 fathers. Refusals were justified by lack of time to participate (n = 3) and psychological unavailability (n = 1). During data collection, 2 mothers were missed due to medical complications and 31 fathers were absent due to professional commitments or emigration.

Trained interviewers were responsible for conducting face-to-face interviews, 15 to 22 days after birth (Mean (SD) = 17.6 (2.3)), using structured questionnaires, to mothers and fathers, separately. Data on sociodemographic characteristics (gender, age, marital status, education and income), previous pregnancies and previous children (biological/adoptive vs. no children) were collected through self-report. Social support was assessed by the Multidimensional Scale of Perceived Social Support (MSPSS) [24] which measures the perceived adequacy of social support received from a significant other, from family and friends. Afterwards, mothers and fathers were asked to fill the validated Portuguese version of the Parental Stressor Scale: Neonatal Intensive Care Unit [25], a self-administered scale consisting of 26 items designed to measure parental perception of sources of stress arising from the environment of the NICU. Each item ranges from 1 (not at all stressful) to 5 (extremely stressful), being grouped into 3 dimensions: "Sights and Sounds" (6 items), "Baby Looks and Behaves" (13 items) and "Change in Parental Role" (7 items). Also, at the end of the questionnaire there is a question about "Overall stress". The score of each of the abovementioned dimensions of the stress scale is calculated as the mean of the group of the respective individual items. It ranges from 1 to 5, with higher values indicating higher levels of parental stress.

Clinical records were reviewed by interviewers to retrieve data on pregnancy complications (which included infectious, placental, haemorrhagic and cardiovascular complications), multiple pregnancy (yes/no), and mode of delivery (vaginal or instrumental and caesarean). Data on the infant's sex, birth weight and gestational age were also collected. Extremely low birth weight and extremely premature infants were defined as birth weight bellow 1000 g [1] and gestational age under 28 weeks [1], respectively.

Statistical analysis was performed using Stata 11.0 (College Station, TX, 2009). Sample characteristics are presented as counts and proportions. The overall score of each subscale, stratified by gender, was presented as medians and percentiles (P25-P75) and compared using the Mann-Whitney test. Mean differences (β) in sources of stress and the respective 95% confidence intervals (95% CI) adjusted for age, educational level and all variables significantly associated with each subscale, were estimated by multiple linear regression models, stratified by gender.

3. Results

The characteristics of the study participants are summarized in Table 1. Almost 70% of the mothers and >75% of the fathers had 30 or more years of age. Most participants were married and this was the first pregnancy for >50% of them. <40% of the participants stated a household monthly income above 1500€, with approximately 40% of the mothers and 30% of the fathers reporting an educational level above 12 years. Pregnancy complications were described by >40% of mothers and fathers, caesarean was the most frequent mode of delivery and almost a quarter of pregnancies were multiple. Approximately 30% and 20% of pregnancies resulted in an extremely low birth weight delivery and in an extremely premature delivery, respectively.

Mothers of very preterm infants hospitalized in NICU classified the overall experience of hospitalization as very stressful (Median (P25–P75): 4.0 (3.0–5.0)), while fathers perceived such situation as stressful (Median (P25–P75): 3.0 (3.0–4.0)) (Table 2). Despite gender differences in the importance attributed to parental stressors, with mothers quoting significantly higher in all subscales, "change in parental role" was classified as the most stressful both by mothers (Median (P25–P75): 4.1 (3.2–4.7)) and fathers (Median (P25–P75): 3.2 (2.4–4.0)).

Tables 3 and 4 present mothers' and fathers' parental stressor subscales scores according to sociodemographic, obstetric and infants' characteristics, respectively. Women with multiple pregnancies reported

Table 1
Participants' characteristics, according to gender.

	Mothers (n = 120) n (%)	Fathers (n = 91) n (%)
Age (years)		
<30	38 (31.7)	22 (24.2)
≥30	82 (68.3)	69 (75.8)
Marital status		
Married/living with a partner	104 (86.8)	80 (87.9)
Single/divorced/widow	16 (13.3)	11 (12.1)
Education level (years)		
≤12	68 (56.7)	64 (70.3)
>12	52 (43.3)	27 (29.7)
Household income (€/month)		
≤1500	73 (62.4)	56 (61.5)
>1500	44 (37.6)	35 (38.5)
Gravidity		
1	63 (52.5)	54 (59.3)
>1	57 (47.5)	37 (40.7)
Previous children		
No	86 (71.7)	68 (74.7)
Yes	34 (28.3)	23 (25.3)
Pregnancy complications[a]		
No	70 (58.3)	51 (56.0)
Yes	50 (41.7)	40 (44.0)
Mode of delivery		
Vaginal/instrumental	38 (31.7)	27 (29.7)
Caesarean	82 (68.3)	64 (70.3)
Multiple pregnancy		
No	92 (76.7)	69 (75.8)
Yes	28 (23.3)	22 (24.2)
Extremely low birth weight deliveryb		
No	83 (69.2)	65 (71.4)
Yes	37 (30.8)	26 (28.6)
Extremely preterm deliveryc		
No	95 (79.2)	71 (78.0)
Yes	25 (20.8)	20 (22.0)

[a] Infectious, placental, haemorrhagic and cardiovascular complications.
[b] <1000 g.
[c] <28 gestational weeks.

lower levels of stress on "change in parental role" ($p = 0.026$) and "overall stress" ($p = 0.020$). Younger fathers (<30 years) presented higher levels of stress in all subscales and "overall stress". Fathers of an extremely preterm ($p = 0.013$) or an extremely low birth weight infant ($p = 0.002$) also revealed higher levels of "overall stress". The latter also attributed higher stress levels to "baby looks and behaves" ($p = 0.012$). Educational level, previous pregnancies, previous children, pregnancy complications, mode of delivery and extremely preterm delivery had no significant associations with parental stress.

After adjustment, multiple pregnancy remained associated with lower levels of stress regarding "change in parental role" and "overall stress" among mothers, while having 30 or more years of age was found to be a significant predictor for decreased fathers' stress related to all subscales and "overall stress" (Table 5).

Table 2
Parental Stressor Scale subscales scores, according to gender.

Parental Stressor Scale[a]	Mothers Median (P25–P75) n = 120	Fathers Median (P25–P75) n = 91	*p*
Sights and sounds	2.7 (2.2-3.4)	2.5 (1.8-3.0)	**0.004**
Baby looks and behaves	3.3 (2.5-4.2)	2.9 (2.2-3.6)	**0.002**
Change in parental role	4.1 (3.2-4.7)	3.2 (2.4-4.0)	**<0.001**
Overall stress	4.0 (3.0-5.0)	3.0 (3.0-4.0)	**0.004**

Bold figures represent significant differences between mothers and fathers, considering $p < 0.05$.
[a] Higher values indicate higher levels of parental stress (range for each subscale: 1–5).

4. Discussion

This study found that the perception of stress during hospitalization of a very preterm infant in NICU is higher among mothers than fathers. It also highlighted gender differences in the factors associated with the perception of stress: fathers' perception of stress was associated with age, while mothers' perceptions were related with multiple pregnancy. These findings contribute to a growing but still scarce literature addressing the specific factors associated with stress induced by the hospitalization of very preterm infants in NICU. This study also showed that, despite gender differences, "change in parental role" was the highest source of stress both for mothers and fathers, and the whole experience of hospitalization was classified as more stressful than the subscales "baby looks and behaves" and "sights and sounds".

"Change on parental role" was the subcale classified as the most stressful among mothers and fathers in our study. These results are corroborated by previous research conducted with parents of premature and term infants [4,13,14] and may be explained by parents' unability to assume their role as primary caregivers in NICU. This situation may lead them to feel less confident and incompetent in their parental roles due to physical and symbolic barriers that prevent them from touching and holding the child [8], and the feeling that they are unable to protect the child from harm [5], that may arise from a discrepancy between parents' social representation of their child and the real premature infant [6]. Our study also shows that the dimensions of "sights and sounds" and "baby looks and behaves" were less stressful for parents, when compared with "change in parental role". Throughout hospitalization, infant become more stable [26] and parents tend, simultaneously, to develop a mutually trustworthy relationship with health professionals, to feel more active in their child's care and more comfortable at NICU [12,27]. However, they continue to be prevented from performing traditional parental roles which might explain the strength of change in parental role as a predictor of stress among parents of infants hospitalized in NICU.

Gender is described as an important determinant of psychosocial stress and, as evidenced by our study, the literature has consistently showed that women tend to report higher levels of stress than men and to attribute more importance to stressful events [28]. Mothers of hospitalized infants, in particular, are more often exposed to daily stressful circumstances given their role as primary caregivers. They also spend more time in NICU and are more aware of caregiving activities [27,29]. Unlike mothers, who tend to focus mainly on the infant [30], fathers who adopt the traditional role of breadwinner, who is responsible for protecting the whole family [31], may be simultaneously concerned with the child, the mother and the work/external environment. This may have a protective effect regarding fathers' perception of stress. Mothers and fathers also cope differently with stressful conditions or events in NICU: fathers tend to delegate care to mothers, while leading the communication with healthcare professionals and focusing on work as a mechanism for distraction [29,32]; mothers use proximity and active participation in the infant's care as a coping mechanism [26,27,31,32]. Furthermore, cultural beliefs that endorse demonstrations of fearlessness and the appearance of being strong as a means of enacting masculinity [33] may contribute to explain why fathers in our study tended to perceive the experience of their infant's hospitalization as less stressful than mothers. Nevertheless, these findings should be interpreted with some caution. If we look at measures of physiological stress responses, no differences between women and men are observed when they are exposed to the same stressor [34]. This suggests that our results may not necessarily be indicating that fathers have lower perceived stress levels, but rather that measures aiming to assess self-reported stress are not sufficiently sensitive to pick up perceived stress among men in its entirety, perhaps due to the social complexities of gender. Further research on the degree of gender sensitivity of stress measures is needed.

Table 3
Mothers' Parental Stressor Subscales scores, according to sociodemographic, obstetric and infant's characteristics (n = 120).

	Parental Stressor Subscale[a]			
	Sights and sounds median (P25–P75)	Baby looks and behaves median (P25–P75)	Change in parental role median (P25–P75)	Overall stress median (P25–P75)
Age (years)				
<30	2.9 (2.3–3.3)	3.6 (2.9–4.2)	4.3 (3.3–4.8)	4.0 (3.0–5.0)
≥30	2.7 (2.2–3.4)	3.2 (2.5–4.2)	4.0 (3.2–4.6)	4.0 (3.0–5.0)
P	0.601	0.276	0.252	0.748
Marital status				
Married/living with a partner	2.8 (2.2–3.4)	3.3 (2.6–4.1)	4.0 (3.2–4.7)	4.0 (3.0–5.0)
Single/divorced/widow	2.6 (2.2–3.8)	3.5 (2.4–4.5)	4.4 (3.5–4.7)	4.0 (3.0–4.0)
P	0.343	0.597	0.347	0.301
Education level (years)				
≤12	2.7 (2.3–3.3)	3.4 (2.7–4.1)	4.1 (3.3–4.7)	4.0 (3.0–4.0)
>12	2.8 (2.2–3.6)	3.3 (2.4–4.2)	4.0 (3.2–4.6)	4.0 (3.0–5.0)
p	0.599	0.882	0.495	0.116
Household income (€/month)				
≤1500	2.7 (2.3–3.3)	3.4 (2.8–4.1)	4.1 (3.4–4.6)	4.0 (3.0–4.0)
>1500	2.8 (2.3–3.6)	3.4 (2.6–4.3)	4.1 (3.2–4.7)	4.0 (3.0–5.0)
p	0.316	0.915	0.746	0.430
Gravidity				
1	2.8 (2.3–3.5)	3.3 (2.7–4.1)	4.1 (3.4–4.7)	4.0 (3.0–5.0)
>1	2.7 (2.2–3.3)	3.3 (2.5–4.3)	4.0 (3.2–4.7)	4.0 (3.0–4.0)
p	0.404	0.971	0.582	0.318
Previous children				
No	2.8 (2.2–3.5)	3.3 (2.5–4.2)	4.1 (3.3–4.7)	4.0 (3.0–5.0)
Yes	2.7 (2.2–3.3)	3.4 (2.5–4.2)	4.0 (3.1–4.7)	4.0 (3.0–5.0)
p	0.352	0.664	0.912	0.919
Pregnancy complications[b]				
No	2.8 (2.3–3.5)	3.4 (2.6–4.2)	4.1 (3.3–4.7)	4.0 (3.0–4.0)
Yes	2.7 (2.2–3.3)	3.3 (2.5–4.2)	4.0 (3.2–4.7)	4.0 (3.0–5.0)
p	0.203	0.563	0.804	0.596
Mode of delivery				
Vaginal/instrumental	2.8 (2.2–3.3)	3.3 (2.5–4.0)	4.2 (3.4–4.7)	4.0 (3.0–4.0)
Caesarean	2.7 (2.2–3.4)	3.4 (2.5–4.2)	4.0 (3.1–4.7)	4.0 (3.0–5.0)
p	0.625	0.693	0.361	0.680
Multiple pregnancy				
No	2.8 (2.3–3.4)	3.3 (2.8–4.2)	4.1 (3.5–4.7)	4.0 (3.0–5.0)
Yes	2.7 (2.2–3.4)	3.4 (2.3–4.2)	3.5 (2.7–4.5)	3.0 (2.0–4.0)
p	0.469	0.491	**0.026**	**0.020**
Extremely low birth weight delivery[c]				
No	2.8 (2.2–3.4)	3.3 (2.5–4.1)	4.0 (3.1–4.6)	4.0 (3.0–4.0)
Yes	2.7 (2.2–3.3)	3.8 (2.9–4.4)	4.3 (3.4–4.9)	4.0 (3.0–5.0)
p	0.831	0.173	0.112	0.102
Extremely preterm delivery[d]				
No	2.7 (2.2–3.3)	3.3 (2.5–4.2)	4.0 (3.1–4.7)	4.0 (3.0–4.0)
Yes	3.3 (2.3–3.8)	3.8 (3.2–4.4)	4.3 (3.4–4.8)	4.0 (3.5–5.0)
p	0.166	0.088	0.157	0.125

Bold figures represent significant differences considering $p < 0.05$.

[a] Higher values indicate higher levels of parental stress (range for each subscale: 1–5).

[b] Infectious, placental, haemorrhagic and cardiovascular complications.

[c] <1000 g.

[d] <28 gestational week.

The differences between the factors associated with parents' perception of stress in NICU identified by this study offer a good opportunity to discuss multiple femininities and masculinities. Studies assessing the perception of stress by mothers of infants admitted to NICU show contradictory results. While some found increased stress levels among older [35,36], unmarried, low-income and less-educated mothers, independently of the infants' gestational age [17], others demonstrated that younger mothers, married and more educated reported higher stress levels [4]. In contrast, our study found no significant associations between mothers' stress levels and variables such as age, marital status, income or level of education. However, our results show that a multiple pregnancy is a significant predictor for decreased mothers' stress levels in NICU. As observed in a recent study, mothers with multiple pregnancies tend to feel overburden during pregnancy, given increased medical involvement and physical discomfort, and also to expect neonatal risks, including preterm birth [37]. These mothers may have been better prepared throughout their pregnancy for complications in the neonatal period and therefore for handling the stress of the NICU. Additionally, the stress of the pregnancy, labour and birth experienced by mothers with multiple babies may be so acute that they perceive the stress in the aftermath of a preterm delivery as being relatively easier to cope with.

Our study also shows that age is a significant predictor for decreased fathers' stress levels in NICU: having 30 or more years of age is a protective factor. Although the literature about fathers' perceived stress is scarce, one study notes that older fathers may experience lower levels of stress because they benefit from a broader lack of previous life experiences that are helpful in preparing them for assuming parental roles and responsibilities [38]. However, we could expect mothers to experience a similar process. Perhaps younger men experience additional fears concerning the wellbeing of their partner and infant and the future of their family because they tend to have less security at work and are therefore less likely to resort to their working environment to buffer stress as older men might do.

No significant differences were observed in the experience of stress by socioeconomic factors (education or income) in men and women. This may be linked to the acuteness of a NICU experience where families

Table 4
Fathers' Parental Stressor Subscales scores, according to sociodemographic, obstetric and infant's characteristics (n = 91).

	Parental Stressor Subscale[a]			
	Sights and sounds median (P25–P75)	Baby looks and behaves median (P25–P75)	Change in parental role median (P25–P75)	Overall stress median (P25–P75)
Age (years)				
<30	2.8 (2.5–3.2)	3.6 (3.2–4.2)	3.7 (3.3–4.3)	4.0 (3.0–5.0)
≥30	2.2 (1.7–3.0)	2.7 (2.1–3.3)	3.0 (2.4–3.7)	3.0 (2.0–4.0)
p	**0.014**	**<0.001**	**0.002**	**0.002**
Marital status				
Married/living with a partner	2.5 (1.8–3.2)	3.0 (2.3–3.5)	3.2 (2.5–3.9)	3.0 (3.0–4.0)
Single/divorced/widow	2.2 (1.8–2.6)	2.6 (2.2–3.7)	3.4 (2.1–4.7)	3.0 (2.0–4.0)
p	0.344	0.597	0.347	0.301
Education level (years)				
≤12	2.5 (1.8–3.1)	3.0 (2.3–3.6)	3.4 (2.4–4.0)	3.0 (2.0–4.0)
>12	2.5 (1.8–2.8)	2.8 (2.1–3.3)	3.0 (2.5–3.6)	3.0 (3.0–4.0)
p	0.600	0.343	0.193	0.774
Household income (€/month)				
≤1500	2.5 (1.8–3.2)	2.9 (2.2–3.6)	3.3 (2.4–4.0)	3.0 (2.0–-4.0)
>1500	2.5 (1.8–3.0)	3.1 (2.3–3.5)	3.1 (2.6–3.9)	3.0 (3.0–4.0)
p	0.316	0.915	0.746	0.430
Gravidity				
1	2.4 (1.8–3.0)	2.8 (2.1–3.4)	3.3 (2.5–4.0)	3.0 (3.0–4.0)
>1	2.5 (1.7–3.2)	3.1 (2.5–4.0)	3.0 (2.4–3.9)	3.0 (3.0–4.0)
p	0.585	0.108	0.862	0.849
Previous children				
No	2.5 (1.8–3.0)	2.9 (2.2–3.5)	3.3 (2.5–4.0)	3.0 (3.0–4.0)
Yes	2.5 (1.7–3.4)	3.1 (2.4–4.2)	3.0 (2.4–3.9)	3.0 (3.0–4.0)
p	0.938	0.281	0.583	0.538
Pregnancy complicationsa				
No	2.6 (1.8–3.0)	3.0 (2.4–3.5)	3.3 (2.5–4.0)	3.0 (3.0–4.0)
Yes	2.4 (1.7–3.1)	2.9 (2.1–3.7)	3.0 (2.4–3.9)	3.0 (2.0–4.0)
p	0.496	0.767	0.440	0.322
Mode of delivery				
Vaginal/instrumental	2.5 (1.8–3.0)	2.9 (2.1–3.4)	3.1 (2.4–3.7)	3.0 (3.0–4.0)
Caesarean	2.5 (1.8–3.2)	2.9 (2.2–3.7)	3.2 (2.5–4.0)	3.0 (3.0–4.0)
p	0.767	0.404	0.478	0.857
Multiple pregnancy				
No	2.5 (1.8–3.0)	2.9 (2.3–3.6)	3.0 (2.5–4.0)	3.0 (3.0–4.0)
Yes	2.4 (1.7–3.2)	2.9 (2.0–3.5)	3.4 (2.4–3.9)	3.0 (2.0–4.0)
p	0.959	0.562	0.860	0.059
Extremely low birth weight deliveryb				
No	2.5 (1.8–2.8)	2.7 (2.1–3.4)	3.2 (2.4–3.9)	3.0 (2.0–4.0)
Yes	2.8 (1.8–3.2)	3.4 (3.0–3.8)	3.1 (2.7–4.0)	4.0 (3.0–5.0)
p	0.262	**0.012**	0.547	**0.002**
Extremely preterm deliveryc				
No	2.3 (1.8–2.8)	2.8 (2.1–3.5)	3.3 (2.4–4.0)	3.0 (2.0–4.0)
Yes	2.9 (1.7–3.3)	3.4 (2.5–4.1)	3.0 (2.8–3.8)	4.0 (3.0–5.0)
p	0.240	0.073	0.916	**0.013**

Bold figures represent significant differences considering p < 0.05.

[a] Infectious, placental, haemorrhagic and cardiovascular complications.

[b] <1000 g.

[c] <28 gestational weeks.

[a] Higher values indicate higher levels of parental stress (range for each subscale: 1–5).

Table 5
Adjusted mean difference (β) of the mothers' and fathers' Parental Stressor Scale scores, according to sociodemographic, obstetric and infant's characteristics.

	Sights and sounds	Baby looks and behaves	Change in parental role	Overall stress
	Adjusted β (95% CI)[a]	Adjusted β (95% CI)[a]	Adjusted β (95% CI)[a]	Adjusted β (95% CI)[a]
Mothers				
Age, years (≥30 vs. <30)	−0.120 (−0.438 to 0.198)	−0.188 (−0.593 to 0.216)	−0.120 (−0.509 to 0.269)	0.001 (−0.413 to 0.414)
Education level, years (≤12 vs >12)	0.116 (−0.183 to 0.414)	0.006 (−0.374 to 0.385)	−0.083 (−0.449 to 0.283)	0.327 (−0.063 to 0.717)
Multiple pregnancy (Yes vs. No)	–	–	**−0.597 (−1.020 to −0.174)**	**−0.603 (−1.052 to −0.153)**
Fathers				
Age, years (≥30 vs. <30)	**−0.510 (−0.919 to −0.101)**	**−0.794 (−1.238 to −0.351)**	**−0.670 (−1.099 to −0.240)**	**−0.756 (−1.290 to −0.222)**
Education level (≤12 vs >12)	−0.025 (−0.408 to 0.359)	−0.205 (−0.612 to 0.203)	−0.240 (−0.642 to 0.162)	0.014 (−0.467 to 0.495)
Extremely low birth weight delivery (Yes vs. No)	–	0.358 (−0.060 to 0.775)	–	0.537 (−0.003 to 1.077)
Extremely preterm delivery (Yes vs. No)	–	–	–	0.440 (−0.130 to 1.010)

95% CI, 95% confidence interval; β, mean differences.

Bold figures represent significant differences, taking into account the interpretation of the 95%CI that should not include the value zero to be statistically significant.

[a] Adjusted for age and educational level and all statistically significant variables for each subscale.

tend to find themselves in the same situation, independently of their social backgrounds. However, once the child returns home, this experience of stress may become differentiated based on factors such as socioeconomic position or social support [39]. A future longitudinal follow-up study is necessary to elucidate this question.

Methodologically, this study offers two major contributions. First, it includes fathers in the research. This is a key methodological advantage both because it enabled the identification of gender differences in the perception of stress and an examination of the factors associated with these differences. Another major contribution relates to the comparison of stress levels between mothers and fathers of very preterm infants and their association with sociodemographic, obstetric and infant's characteristics in a representative sample: data collection was carried out over an extended recruitment period of one year and participants were consecutively and systematically invited to participate in all level III NICU from the North of Portugal. Despite the innovative nature of the present study, some limitations should be discussed. First, the exploratory results require replication. It is also important to highlight the need to compare the perception of stress among parents of very preterm infants with the perceptions of parents with non-very preterm infants hospitalized in NICU. Additionally, further studies comparing stress scores according to regimes of family-centered care and parental feelings of self-blame and guilt related to birthing a preterm infant are needed. Furthermore, it is crucial to identify potential stressors beyond the NICU environment, including stressful family environments and stress experienced within couples.

One final consideration regards the stress scores identified by our study: when compared to most previous studies about stress among parents of preterm infants and term infants with low birth weight hospitalized in NICU, our study found higher scores on the stress subscales for both mothers and fathers [10,17]. However, it is important to highlight that the studies available report mean scores, and not median scores, which hinders the direct comparison with our results.

Lack of social support did not explain this tendency in the present study, since participants reported high levels of support received from a significant other, family and/or friends and, as a result, it was not significantly associated with parental stress (data not shown). These differences may thus be explained by methodological issues, namely unrepresentative sampling and the moment of data collection, which in some studies took part during the second week of hospitalization [10]. They may also be due to societal issues. Wide differences exist between (and sometimes within) countries in terms of political frameworks on parental leave, regimes of family-centered care, stigmatization and social prejudices associated with prematurity and parental feelings of self-blame and guilt [6] related to birthing a preterm infant. Less favorable environments and circumstances, as it appears to have been the case with the parents included in our study [40] such as absence of parental leave equal to the period of hospitalization for both parents, as well as the lack of privacy in the NICU, are likely to be conducive to more stressful experiences with infant hospitalization. If such high levels of stress were to persist over time and become an integral part of the parenting style and family functioning, major problems related to chronic stress may arise [19].

To conclude, our findings confirm that becoming a parent of a very preterm infant and his/her subsequent hospitalization in NICU is an event associated with emotional distress. Assessing parental sources of stress is essential to promoting parental support and guidance, and to increasing parents' awareness of the key role they can play in the early stages of their child's development. Our findings indicate that it might be advisable for healthcare staff to pay particular attention to younger fathers. This study also points out the need to deliver follow-up care to parents to help them cope with stress, and to consider other factors that may be positively associated with parental stress during hospitalization in NICU, namely lack of social and governmental support, financial and work concerns, stigmatization and social prejudices, and limited attention to family-centered care. Attending to these factors and implementing interventions focused on reducing parental stress is crucial to diminish disparities in family health across and within countries.

Finally, considering that parenting a very preterm infant during hospitalization in NICU will most likely be a stressful event, it is necessary to assess its potential damage in the long-term and to explore how it may affect family functioning and wellbeing.

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4.2. PAPER III: A short form of the neonatal intensive care unit family needs inventory

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ORIGINAL ARTICLE

A short form of the neonatal intensive care unit family needs inventory[☆,☆☆]

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Neonatal intensive care units;
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Abstract

Objective: The identification of parental needs in Neonatal Intensive Care Units is essential to design and implement family-centered care. This article aims to validate the Neonatal Intensive Care Units Family Needs Inventory for the Portuguese population, and to propose a Short Form.

Methods: A linguistic adaptation of the Neonatal Intensive Care Units Family Needs Inventory, a self-report scale with 56-items, was performed. The instrument was administered to 211 parents of infants hospitalized in all level III Neonatal Intensive Care Units in the North of Portugal, 15-22 days after admission (July of 2013-June of 2014). The number of items needed to achieve reliability close to 0.8 was calculated using by the Spearman-Brown formula. The global goodness of fit of the scale was evaluated using the comparative fit index. Construct validity was assessed through association of each dimension score with socio-demographic and obstetric characteristics.

Results: Exploratory factor analysis revealed two dimensions, one focused on parents' needs and another on the infant's needs. To compose the Short Form Inventory, items with ceiling effect were eliminated and 22 items were submitted to confirmatory analysis, which supported the existence of two dimensions (CFI = 0.925). The Short Form showed a high degree of reliability (alpha $\geq$ 0.76). Less educated and older parents more frequently attributed a significantly higher importance to parent-centered needs, while parents of multiples revealed a tendency to value infant-centered needs.

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Conclusions: The Short Form of the Neonatal Intensive Care Units Family Needs Inventory is a brief, simple, and valid instrument with a high degree of reliability. Further studies are needed to explore associations with practices of family-centered care.

PALAVRAS-CHAVE
Unidade de Cuidados Intensivos Neonatais;
Avaliação de necessidades;
Pais;
Estudos de validação como assunto

Versão curta do inventário de necessidades da família na unidade de cuidados intensivos neonatais

Resumo

Objetivo: A identificação de necessidades parentais em Unidades de Cuidados Intensivos Neonatais (UCINs) é essencial para planejar e implementar cuidados centrados na família. Este artigo pretende validar o Inventário de Necessidades da Família em UCIN na população portuguesa e propor uma Versão Curta do mesmo.
Metodologia: Foi realizada uma adaptação linguística do Inventário de Necessidades da Família na UCIN, uma escala de autorrelato com 56 itens. O instrumento foi aplicado a 211 pais de bebês internados em todas as UCINs de nível III no Norte de Portugal 15 a 22 dias após a internação (julho de 2013-junho de 2014). O número de itens necessários para atingir uma confiabilidade próxima a 0,8 foi calculado pela fórmula de Spearman-Brown. A adequação global da escala foi avaliada pelo índice de ajuste comparativo (CFI). A validade de construto foi avaliada através da associação do escore de cada dimensão com características sociodemográficas e obstétricas.
Resultados: A análise fatorial exploratória revelou duas dimensões, uma focada nas necessidades dos pais e outra, nas necessidades do bebê. Para compor a Versão Curta do Inventário, os itens com efeito teto foram eliminados, e 22 itens foram submetidos a análise confirmatória, que sustentou a existência de duas dimensões (CFI = 0,925). A Versão Curta apresentou alto grau de confiabilidade (alfa $\geq$ 0,76). Pais mais velhos e com menor escolaridade atribuíram, mais frequentemente, maior importância à subescala de necessidades centradas nos pais enquanto os pais de gêmeos revelaram uma tendência de valorizar as necessidades centradas nos bebês.
Conclusões: A Versão Curta do Inventário de Necessidades da Família em UCIN é um instrumento breve, simples e válido com alto grau de confiabilidade. São necessários estudos adicionais para explorar as associações com práticas de cuidados centrados na família.

Introduction

The improvement in the quality of antenatal and neonatal care led to a decrease in perinatal mortality and morbidity in the last three decades,[1] but prematurity is still the worldwide leading direct cause of neonatal death and short- and long-term morbidity.[2] The hospitalization of a newborn in a neonatal intensive care unit (NICU) constitutes a disruptive life event with impact on family health.[3,4] During the hospitalization period, parents need to assume new roles under adverse conditions,[5] such as physical separation from the child, structured and controlled opportunities of interaction, difficulties in feeling part of the infant's care, and fear for his/her survival and future development, while also experiencing feelings of hope, love, and happiness.[3,6,7] Parents have reported the importance of obtaining information and guidance,[8] trusting in the healthcare team,[9] and experiencing support from staff members and from his/her partner.[9,10]

Family-centered care, defined as provision of care that is respectful of and responsive to parents preferences, needs, and values, is essential for a successful design and implementation of evidence-based practices in NICU.[11] Therefore, the identification of parental needs may contribute for diminishing the risk for the development of parental stress and several psychopathological symptoms.[9,12]

The NICU Family Needs Inventory is a reliable self-report instrument to assess the parents' needs during an infant's hospitalization in NICU.[13] To listen to parents is particularly important in a context where healthcare professionals and parents tend to identify different parental needs.[14] To the best of the authors' knowledge, the NICU Family Needs Inventory is the only scale specifically designed and validated for the NICU setting, but its utility for health research, as well as for counseling and clinical practice, is limited by its length (56-items). A shorter version of the Inventory, brief and easy to administrate, but also valid and with a high degree of reliability, would actively contribute to the identification of the specific needs of each family, and for their inclusion in health care, while lessening the intrusion and ensuring the ethical principles of respect for autonomy, non-maleficence, beneficence, and justice.[15]

Thus, this study aimed to validate the Neonatal Intensive Care Unit (NICU) Family Needs Inventory for the Portuguese population, and to propose a Short Form.

Methods

Original instrument

The NICU Family Needs Inventory[13] was the instrument used for data collection. It is a self-reported scale divided into five subscales: support, information, comfort, assurance, and proximity. It consists of 56 need statements designed to measure the importance attributed to family needs by mothers and fathers in the NICU setting, ranging from 1 to 4 (not important, slightly important, important, and very important, respectively). Each item could also be classified as not applicable, if the participants had never experienced the specific need. Permission to use or modify the inventory was granted by the original author.

Linguistic adaptation

Two native Portuguese speakers proficient in English translated NICU Family Needs Inventory and an expert committee, with backgrounds in Public Health, Psychology, and Sociology, ensured conceptual and item equivalence. The discrepancies between the two translations were solved by consensus and merged into a single Portuguese version. After completing the Inventory, parents were invited to report eventual doubts and suggestions.

Sample and recruitment

The adapted version of the Inventory was administered to a cohort of parents of very preterm infants, which has been previously described.[16] Briefly, all parents of very preterm infants born between July 1, 2013 and June 30, 2014 who were hospitalized in all level III NICU that provided subspecialty care for critically ill newborn infants[17] in the Northern Health Region of Portugal ($n = 7$), were consecutively invited to participate in the study. Parents were approached during their hospital stay by a NICU healthcare professional (neonatologist or nurse), who was responsible for the presentation of the study and invitation to participate, 15–22 days after birth. Only parents with infants born before 32 weeks of gestation and still hospitalized in NICU at the time of the interview were considered eligible to participate in the study. Parents with serious illness that precluded NICU visitation (*e.g.* severe chronic conditions), families who were absent from NICU during the hospitalization period, and parents whose infants were discharged or transferred to another hospital were excluded. Among the 126 eligible couples, 122 (96.8%) agreed to participate. The final sample included 120 mothers and 91 fathers (in 89 cases, both parents participated in the study). Refusals were justified by lack of time to participate ($n = 3$) and psychological unavailability ($n = 1$). During data collection, two mothers were absent due to medical complications and 31 fathers were absent due to professional commitments or emigration.

Trained interviewers were responsible for interviewing mothers and fathers, separately. Data on socio-demographic characteristics were collected through face-to-face interviews, using a structured questionnaire. Afterwards, the parents were asked to fill the NICU Family Needs Inventory.[13] Clinical records were reviewed to retrieve data on multiple pregnancy, gravidity, and pregnancy complications (which included infectious, placental, hemorrhagic, and cardiovascular complications). Data on neonatal birth weight and gestational age were also collected. According to the World Health Organization definitions, extremely low birth weight and extremely premature infants were defined as those with birth weight below 1000 g and gestational age under 28 weeks, respectively.[18]

The present investigation was approved by the Ethics Committee of the seven Hospitals with NICU where data was collected and by the National Committee for Data Protection. Each participant signed an informed consent.

Statistical analysis

Due to the low variability in the response scale, items were dichotomized as very important (scoring 4) or not very important (scoring 3 or less, combining the answers not important, slightly important, and important) for analysis. Exploratory factor analysis for dichotomous variables was performed to assess the dimensionality of the scale, whether the construct study is a single concept or multiple concepts. When items are used to form scale they should all be correlated with one another, and they should have internal consistency. Cronbach's alpha was used to measure internal consistency and a Cronbach alpha value of 0.8 or higher was considered as good internal consistency.

Considering that the aim of the study was to construct a short-form, the Spearman–Brown formula[19] allowed for the estimation of the minimum number of items needed to achieve reliability close to 0.8. Items with the best item-fit statistics for the one-parameter logistic model (Rasch model) were included in the Short Version. This ensured that items with similar psychometric quality were selected. The global goodness of fit of the Short Version was evaluated using the comparative fit index (CFI), the Tucker–Lewis Index (TLI), and the root mean squared error approximation (RMSEA), obtained from the confirmatory factor analysis. Cut off values above 0.9 for CFI and TLI, and below 0.10 for RMSEA indicated a good fit of the model.[20]

To assess the construct validity of the Short Version, the authors measured the association of each dimension score with socio-demographic and obstetric characteristics of the participants, and a multilevel model with a random effect by couple was used to estimate the differences between groups, correcting for the couple dependency. It was assumed that those with more adverse social environments would be more likely to attribute higher importance to parents-centered needs. The subscales scores of the Short Version were calculated as the sum of all items after dichotomization. When items were classified as non-applicable, the score was calculated as the mean of all items answered multiplied by the total number of items in each subscale ($n = 10$ for the parents-centered needs subscale and $n = 12$ for the infant-centered needs subscale).

Statistical analyses were performed using the software STATA 11.0 (StataCorp. 2009. Stata Statistical Software: Release 11. College Station, TX, USA), R 2.12.1 (R Statistical Programming Language 2.12.1), and MPlus (MPlus version 5.2.).

Results

The majority of the participants had less than 35 years of age (70.1%), 63.0% had more than nine years of education, and 65.4% reported a household monthly income above € 1000. This was the first pregnancy for 55.5% of the participants and 42.7% had a pregnancy complication. In this sample, 29.9% of the participants had an extremely low birth weight delivery, 21.3% an extremely preterm delivery, and 23.7% of the pregnancies were multiple (Table 1).

Exploratory factor analysis of the NICU Family Needs Inventory

Overall, 60.7% of the need statements were identified as very important needs (mean score > 3.5) by the participants and none of the statements were ranked as not important. Only one item (1.8%), ''to have a pastor, clergy, or

Table 1 Associations between the score of each subscale of the Short Version with socio-demographic and obstetric characteristics of the participants ($n = 211$).

	Total *n* (%)	Parents-centered needs Mean (SD)	*p*	Infant-centered needs Mean (SD)	*p*
Overall		3.5 (2.6)		8.6 (2.7)	
Sex					
Female	120 (56.9)	3.7 (2.6)		8.8 (2.7)	
Male	91 (43.1)	3.3 (2.4)	0.239	8.3 (2.7)	0.230
Age (years)					
<35	148 (70.1)	3.3 (2.3)		8.5 (2.6)	
≥35	63 (29.9)	4.1 (3.0)	0.037	8.8 (2.9)	0.516
Education (years)					
≤ 9	78 (37.0)	4.3 (2.5)		8.7 (2.9)	
> 9	133 (63.0)	3.1 (2.5)	<0.001	8.5 (2.6)	0.600
Household monthly income (€)					
≤1000	72 (34.6)	3.9 (2.6)		8.4 (3.0)	
>1000	136 (65.4)	3.4 (2.6)	0.149	8.7 (2.6)	0.518
Gravidity					
0	117 (55.5)	3.7 (2.6)		8.8 (2.6)	
≥1	94 (44.5)	3.4 (2.5)	0.446	8.2 (2.9)	0.111
Pregnancy complications[a]					
Yes	90 (42.7)	3.4 (2.7)		8.3 (2.9)	
No	121 (57.4)	3.6 (2.5)	0.530	8.8 (2.6)	0.256
Multiple pregnancy					
Yes	50 (23.7)	4.0 (2.6)		9.4 (2.8)	
No	161 (76.3)	3.4 (2.5)	0.117	8.3 (2.7)	0.024
Extremely premature delivery[b]					
Yes	45 (21.3)	3.5 (2.8)		8.4 (3.0)	
No	166 (78.7)	3.6 (2.5)	0.943	8.6 (2.7)	0.692
Extremely low birth weight delivery[c]					
Yes	63 (29.9)	3.3 (2.5)		8.4 (2.6)	
No	148 (70.1)	3.6 (2.6)	0.375	8.6 (2.8)	0.547

SD, standard deviation.

Note: The subscales scores were calculated as the sum of all items after dichotomization as very important or not very important (combining the answers not important, slightly important and important). In the case of having items classified as non-applicable, the score was calculated as the mean of all items answered multiplied by the total number of items in each subscale ($n = 10$ for parents-centered needs subscale and $n = 12$ for infant-centered needs subscale).

[a] Includes the following complications: gestational hypertension; preeclampsia; eclampsia; HELLP syndrome; gestational diabetes mellitus; acute pyelonephritis; metrorragia; placenta previa; placental abruption; fetal malformations; pulmonary infection; urinary tract infections; threat of miscarriage; and cholestasis of pregnancy.

[b] <28 gestational weeks.

[c] <1000 g

Table 2 Proportion of participants who answered ''very important'' in each item, standardized factor loadings for two factors in exploratory factor analysis (EFA).

	Very important *n* (%)	Two factor EFA	
		std.z1	std.z2
Item			
1	191 (91.4)	0.673	−0.173
2	147 (72.1)	0.365	0.196
3	157 (75.5)	0.652	−0.173
4	178 (84.8)	0.658	0.137
5	200 (95.7)	0.773	−0.099
6	186 (91.6)	0.723	−0.022
7	139 (66.2)	0.644	−0.065
8	96 (45.7)	0.389	0.402
9	96 (49.5)	0.319	0.460
10	189 (90.0)	0.848	−0.094
11	181 (86.6)	0.754	−0.116
12	104 (50.0)	0.454	0.288
13	167 (80.7)	0.450	0.255
14	197 (94.7)	0.994	0.651
15	63 (31.3)	0.440	0.358
16	131 (63.6)	0.636	−0.039
17	152 (73.8)	0.493	0.094
18	139 (67.5)	0.701	0.006
19	101 (49.3)	0.262	0.328
20	186 (89.0)	0.902	−0.424
21	56 (27.3)	0.404	0.455
22	22 (11.7)	−0.112	0.941
23	120 (57.7)	0.643	0.081
24	35 (17.7)	−0.009	0.884
25	142 (68.3)	0.614	0.265
26	114 (54.6)	0.546	0.315
27	101 (45.6)	0.557	0.407
28	96 (45.9)	0.482	0.422
29	100 (48.1)	0.312	0.272
30	90 (43.3)	0.221	0.694
31	106 (56.1)	0.274	0.290
32	110 (53.1)	0.411	0.407
33	98 (47.3)	0.317	0.528
34	169 (82.0)	0.595	0.272
35	196 (94.2)	0.824	0.021
36	173 (83.6)	0.650	0.104
37	158 (79.0)	0.612	0.266
38	191 (91.8)	0.893	0.000
39	201 (96.6)	1.008	−0.023
40	192 (92.3)	0.948	−0.348
41	92 (44.4)	0.028	0.835
42	160 (78.8)	0.747	0.094
43	59 (52.2)	0.621	0.358
44	93 (46.0)	0.494	0.445
45	71 (60.2)	0.619	0.341
46	131 (64.5)	0.637	0.208
47	91 (46.4)	0.415	0.507
48	174 (84.1)	0.698	0.268
49	183 (89.7)	0.632	0.153
50	157 (76.6)	0.569	0.081
51	44 (22.1)	0.306	0.467
52	195 (96.1)	0.806	−0.037
53	195 (95.6)	0.967	0.015
54	167 (81.9)	0.460	0.092
55	176 (87.6)	0.651	−0.038
56	195 (95.1)	0.784	0.095

Note: Total of participants may not add 211 in each item due to the non-applicable response option. The items included in the dimension focusing on infant-centered needs are presented at bold.

other person from my church visit'', was regarded as slightly important (mean score between 1.5 and 2.5).

Exploratory factor analysis for dichotomous variables revealed two dimensions, with almost all of the items associated to needs centered on the parents in the first dimension, and all those associated with infant-centered needs in the second (Table 2).

Short Form of the NICU Family Needs Inventory

To compose the Short Form Inventory, all the items with ceiling effect (more than 90% of participants answered ''very important'') were eliminated, which corresponded to items 1, 5, 14, 35, 38, 39, 40, 52, 53, and 56 of the original scale (Table 3). According to the Spearman–Brown prediction formula, 10 and 12 items were included in the dimensions focusing on parents-centered needs and infant-centered needs, respectively. The items with a better fit within dimension 1 were selected, taking into account the item-fit statistics for the Rasch model (Appendix A).

Confirmatory factor analysis supported that both dimensions were reflected in the Short Form version of the Inventory (Fig. 1). Overall, the Short Form NICU Family Needs Inventory showed a high degree of reliability, with a Cronbach's alpha of 0.77 for the parents-centered needs dimension and 0.76 for the infant-centered needs dimension. A significant correlation was observed between the two dimensions, indicating a considerable dependence between the subscales. The CFI, TLI, and RMSEA values obtained from confirmatory factor analysis indicated an adequate goodness of fit in this study sample (X^2 (50) = 643.690; CFI = 0.925; TLI = 0.950; RMSEA = 0.053).

The five subscales of the original Inventory (assurance, proximity, information, comfort, and support) were reorganized within the two new dimensions obtained in the Short Form version. Support and comfort needs were mainly included in the parents-centered needs subscale, while information, assurance, and proximity were predominantly included in the infant-centered needs subscale (Table 3).

Associations between each subscale of the Short Version score with socio-demographic and obstetric characteristics of the participants are presented in Table 1. Parents of infants hospitalized in NICU classified, on average, the infant-centered needs subscale as more important than the parents-centered needs subscale (mean (SD): 8.6 (2.7) [range: 0–12]; 3.5 (2.6) [range: 0–10], respectively). Overall, a high concordance was observed in the perception

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Note: The Appendix A of the paper can be consulted in the Appendix 3 of the thesis.

Table 3 Presentation of the items composing the Portuguese Short Version of the Neonatal Intensive Care Unit (NICU) Family Needs Inventory.

Item number Short Version	Item number NICU Family Needs Inventory	Previous subscale	Short-Form dimension
1	3	Proximity	Infant-centered needs
2	4	Information	Infant-centered needs
3	8	Comfort	Parents-centered needs
4	11	Information	Infant-centered needs
5	13	Information	Infant-centered needs
6	15	Support	Parents-centered needs
7	16	Information	Infant-centered needs
8	19	Support	Parents-centered needs
9	20	Information	Infant-centered needs
10	21	Comfort	Parents-centered needs
11	22	Support	Parents-centered needs
12	24	Comfort	Parents-centered needs
13	25	Comfort	Infant-centered needs
14	27	Support	Infant-centered needs
15	29	Proximity	Infant-centered needs
16	31	Support	Parents-centered needs
17	32	Comfort	Infant-centered needs
18	33	Information	Parents-centered needs
19	36	Information	Infant-centered needs
20	47	Support	Parents-centered needs
21	50	Assurance	Infant-centered needs
22	51	Support	Parents-centered needs

of needs between couples for the infant-centered needs subscale (35.0%), while the concordance regarding the parents-centered needs subscale was much lower (14.3%).

Parents with more than 34 years of age and with less than ten years of education were significantly more likely to attribute a higher importance to the subscale focusing on the parents' needs than those younger and more educated (mean (SD): 3.3 (2.3) *vs.* 4.1 (3.0), $p = 0.037$; mean (SD): 4.3 (2.5) *vs.* 3.1 (2.5), $p < 0.001$). Also, parents of multiples revealed a significant tendency to value infant-centered needs, in comparison with those without a multiple pregnancy (mean (SD): 9.4 (2.8) *vs.* 8.3 (2.7), $p = 0.024$).

Discussion

In the present study, the authors propose a Short Form of the NICU Family Needs Inventory, a brief and valid instrument for the Portuguese population. It is composed by 22 items divided into two subscales, one focusing on parents-centered needs and another on infant-centered needs. The Short Form Inventory presented a high degree of reliability to assess the parental needs of parents during their child's hospitalization in NICU, and had a high internal consistency in both dimensions. Also, an adequate goodness of fit ensured that the model fit the data analyzed well.

Overall, all items of the Short Form of the Inventory were highly valued by the parents, underlining the relevance of those issues for the design of parent-friendly NICU settings and highlighting the importance of family-centered care in such a context.[11] In fact, previous studies concluded that, during the hospitalization period, parents face particular infant-centered needs, such as need of information,[6,8] assurance,[9,10] and proximity[6] that intertwine with their own needs of comfort[10] and social support.[9,10] Access to accurate and consistent information, the need to trust in the healthcare team, as well as, assuming responsibility for the infant, by comforting, visiting, breastfeeding, bathing, diapering, and touching the child, increase parents' sense of control and empowerment, contributing to their involvement in infant's health and care by focusing on their child.[21,22] At the same time, establishing genuine relationships with the staff, relying on their partner for physical help and emotional support, making the environment more homelike, and the possibility of accommodation at bedside improve parents' satisfaction with the healthcare[23] and contribute for their health and well-being by focusing on their own needs.[24,25]

The four categories of response of the original instrument (not important, slightly important, important and very important) must be included in the Short Form administration sheet, in order to allow a higher variability of responses and a better discrimination of the parental perception of needs in NICU. However, this proposal suggests the dichotomization of the scores into not very important and very important for purposes of data analysis, which facilitates the interpretation of the results.

This is, to the best of the authors' knowledge, the only reliable and valid instrument available to evaluate parental needs during infants' hospitalization in NICU and there is no gold standard for this construct. Therefore, criterion validity was not assessed in the present study, since it was not possible to compare the results with an existing instrument

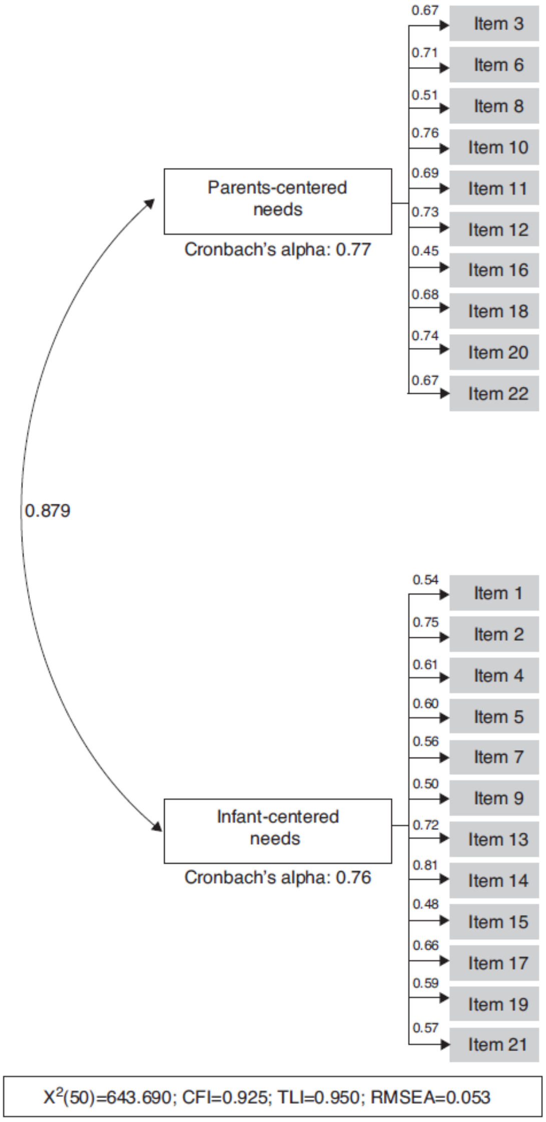

Figure 1 Confirmatory factor analysis of the Portuguese Short Version of the Neonatal Intensive Care Unit (NICU) Family Needs Inventory.

or an external and objective measure of the construct.[26] The strategy implemented for assessing construct validity consisted in measuring the association of each dimension score with socio-demographic and obstetric characteristics of the participants. The authors concluded that older and less educated parents were significantly more likely to attribute higher importance to parents-centered needs, suggesting the re-entrenchment of social inequalities inside NICU. It highlights how social factors, namely socioeconomic position, impact on valuing parental needs during hospitalization of their infants.

Also, mothers attributed higher importance to both parents and infant needs, although not reaching statistical significance. These results were expected since mothers frequently take over the care of the child during hospitalization.[9] In fact, the idea of intensive motherhood, in which mothering is exclusive, wholly child centered, emotionally involving, and time consuming,[27] is reinforced in NICU. The assumption that mothers, more than fathers, should be devoted to child care and self-sacrifice,[28] through the naturalization and devaluation of the pain and suffering during hospitalization, might have justified the predominance of infant-centered needs over parents-centered needs. These results emphasize the previously described focus on infants' health and prognosis over the fulfillment of parents' needs.[29]

A major strength of this study is the proposal of a brief and easy to administrate instrument, simple to score and to interpret. Although most of parents have time available to answer to the full inventory, this does not mean that they have the emotional availability to do so. A very preterm delivery and hospitalization in NICU leads to an emotional chaos in which parents frequently report feelings of fear, sadness, guilt, or failure.[5] Considering parents' emotional availability is particularly relevant in a context where healthcare professionals and researchers must act according to fundamental ethical principles, including the minimization of the burden and the decrease of the intrusion into the private lives of parents of children hospitalized in NICU. Therefore, the Short Form of the Inventory can contribute to increase parental participation and to minimize the burden, while allowing for the identification and inclusion of family needs in healthcare, taking into account essential ethical principles.[15]

Some limitations of the present study should be acknowledged and discussed. Although the sample size was relatively small for a factor analysis, it has been argued that the increase in precision gained from sample sizes over 50 is rarely worth the effort.[30] Also, data were collected from all level III NICU located in the North of Portugal, and the proportion of refusals was very low, ensuring the representativeness of the sample.

The Short Form Inventory demonstrated good psychometric properties. However, further studies comparing the performance of the instrument with the original one, according to the parents' socio-demographic characteristics and reproductive and obstetric history, as well as to infants' characteristics at birth and prognosis, are needed. Furthermore, it is important to determine its robustness and reliability in different cultural, economic, and political contexts.

In conclusion, the Short Form of the NICU Family Needs Inventory is a valid and reliable instrument to measure parents-centered and infant-centered needs, by combining the five subscales of the original Inventory. This shorter version of the Inventory, valid for the Portuguese population, is brief and easy to administrate.

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Conflicts of interest

The authors declare no conflicts of interest.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.jped.2015.04.010.

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4.3. PAPER IV: Needs of parents of very preterm infants in Neonatal Intensive Care Units: A mixed methods study

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Submitted

Needs of parents of very preterm infants in Neonatal Intensive Care Units: A mixed methods study

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Abstract

Objective: To explore the needs of parents of very preterm infants hospitalised in Neonatal Intensive Care Units according to their socioeconomic position, obstetric history and infant's characteristics, intending to promote quality healthcare systems and services in neonatology.

Design: Sequential explanatory observational mixed methods study.

Setting: All level III public Neonatal Intensive Care Units in North Portugal.

Participants: Mothers and fathers of very preterm infants hospitalised between July 2013 and June 2014.

Main outcome measures: Family needs of assurance, proximity, information, support and comfort.

Results: Mothers valued more information needs than fathers and their overall scores were mainly influenced by age and educational level, while fathers' perception of their needs was mainly influenced by previous children. Despite gender differences, the assurance and proximity needs of parents apply across sociotechnical environments. Qualitative findings added the following needs: instrumental support from the government; regular emotional support from psychologists and social workers; enhancement of privacy in the neonatology ward to assure family-centred information and comfort; and availability of other parents and health professionals as complementary health mediators in the provision of detailed and coherent information.

Conclusions: The promotion of family-friendly and gender-equality policies is crucial to support quality family-centred and integrated healthcare services in neonatology. This study raises awareness for the need of flexibility and sensitivity in developing conceptual frameworks and instruments to assess parental needs that take notice of socioeconomic position and reproductive trajectories of parents, as well as issues of privacy and regular emotional support in Neonatal Intensive Care Units.

Keywords: Needs Assessment; Parents; Intensive Care Units, Neonatal; Family-integrated care.

Introduction

Time is ripe to identify, understand and address parental needs during infants' hospitalisation in Neonatal Intensive Care Units (NICU) by listening to both mothers and fathers' perspectives (1,2). Addressing parental needs is widely recognised as a keys to guide quality family-centred and integrated healthcare services (3-5), as well as to improve parental quality of life (6). These issues are particularly pertinent for parents of preterm infants due to child's vulnerability, increased risk of morbidities and prolonged LOS (7). The incorporation of parents' perspectives on the organization of care and health governance is especially relevant considering the mismatches between the perceptions of health professionals and parents (8,9), and the differences between the support provided to family-friendly and gender-equality policies in Western and Nordic European countries (10). These phenomena could led to a gap between the care expected by parents and the care actually provided (11), as well as discrepancies between guidelines and families' needs (12).

The literature on parental needs in NICU is mainly based on a qualitative assessment of mothers' experiences and has been focused on dimensions related with shared health and care (e.g., information and communication, assurance, proximity, social support, comfort) (6,13,14), and not with the shared governance for health, limiting the opportunities of co-producing family centred and integrated healthcare systems (15). Parents consistently prioritize infant-centred needs and undervalue parents-centred needs [16], but the scarce existing quantitative studies show that such needs may vary according to parents' socioeconomic characteristics (sex, age, marital status, educational level, income) (1,17,18), their previous experiences in NICU (17), infants' illness trajectories (13) and LOS (18), as well as the design, organizational rules and regulations of each NICU (19). These findings suggest that both parental and infants' characteristics and the environment of NICUs influence the type of needs experienced by parents.

Hence, the comprehensive identification and mapping of both mothers' and fathers' needs simultaneously related to shared care and governance would benefit from a mixed methods approach. This knowledge is crucial for promoting quality healthcare systems and services centred on families and enabling their involvement in co-production of health in neonatology (15). By integrating quantitative and qualitative data, this study aims to explore needs of mothers and fathers of very preterm infants hospitalised in NICU according to their socioeconomic position, obstetric history and infant's characteristics.

Methods

This observational mixed methods study used a sequential explanatory design, whereby the quantitative data were first analysed followed by an interpretation of qualitative data (20). The approach comprised individual quantitative questionnaires during infants' NICU hospitalisation, applied 15 to 22 days after delivery, and qualitative semi-structured couple interviews 4 months after. Clinical records were reviewed by researchers to retrieve data on pregnancy complications, multiple pregnancy, and each infant's gestational age and birth weight. Extremely low birth weight was defined as birth weight bellow 1000g and extremely premature infants were those with gestational age under 28 weeks (21).

Between July 2013 and June 2014, mothers and fathers of very preterm infants admitted to all public level III NICU in Northern Health Region of Portugal (n=7) were consecutively and systematically invited to participate in the study by the healthcare team. Eligible parents were those whose infants survived, were present in NICU during the hospitalisation period, and were able to speak and write in Portuguese (22). Among the 126 eligible couples, 122 (96.8%) agreed to participate in the questionnaire and among the latter, 117 (95.9%) accepted to be contacted for the qualitative interviews.

Questionnaire: participants and data collection

Trained interviewers conducted face-to-face interviews, using structured questionnaires, to mothers and fathers, separately but within the same timeframe. Self- reported data on sociodemographic characteristics (sex, age, educational level, marital status, occupation and subjective social class), as well as the existence of previous children were collected.

Occupations were classified according to the Portuguese Classification of Occupations 2010 (23) and grouped in three categories: upper-white-collar, including executive civil servants, industrial directors and executives, professionals and scientists, middle management and technicians; lower-white-collar, including administrative and related workers, and service and sales workers; and blue-collar, which includes farmers and skilled agricultural, fisheries workers, skilled workers, craftsmen and similar, machine operators, assembly workers and unskilled workers.

Each participant was georeferenced according to home address, using the ArcGIS Online World Geocoding Service and Google Maps, to be matched to the contextual variable: distance in minutes from residence to NICU. The shortest road distance from participant's residence to NICU was calculated using ArcGIS version 10.4.1 and the Network analyst extension. The street network, required to calculate road distances, was provided courtesy of Environmental Systems Research Institute.

Additionally, mothers and fathers filled in the NICU Family Needs Inventory (2), a self-report scale consisting in 56 need statements designed to measure the importance attributed to family needs. Each item ranges from 1 to 4: 1 - Not important, 2 - Slightly important, 3 - Important and 4 - Very important, being grouped into 5 subscales: "Assurance", "Proximity", "Information", "Support" and "Comfort". There is one open-ended question in which parents can describe other needs than those asked in the inventory. The Portuguese version of the inventory has shown a good internal consistency (Cronbach's α = 0.92). After excluding participants with >20% of missing values, 118 mothers and 89 fathers (86 couples) were included in quantitative analysis.

Statistical analysis

Scores of the NICU Family Needs Inventory were calculated as the sum of all items of each subscale divided by the number of items for each subscale. In the case of having ≤20% of items classified as non-applicable or missing values, the scores were calculated as the mean of all items answered in each subscale.

Statistical analysis was performed using Stata 11.0 (College Station, TX, 2009). Sample characteristics are presented as counts and proportions and compared by Chi-square Test. The overall score of each subscale is presented as medians and interquartile range (IQR), according to socioeconomic, obstetric and infant's characteristics, and the comparison between mothers and fathers were computed using the Mann-Whitney test.

Qualitative interviews: participants and data collection

Semi-structured qualitative interviews were conducted with a subsample of 26 parental couples between November 2013 and April 2014. Participants were purposively sampled to include parents of infants with extremely and non-extremely low birth weight. In addition, a heterogeneity sampling was used for maximum variation of views and experiences, until no new themes emerged from interview data – thematic saturation (24).

Interviews were conducted at parents' homes (n=19), at the university department responsible for the study (n=6) and in a private hospital room (n=1). Interview duration ranged from 20 to 72 minutes (mean: 39 minutes). All interviews were audio taped and transcribed verbatim and accuracy has been checked. The interview guide covered the following areas: how parents deal with uncertainty and doubts and how they made their decisions concerning parental care, treatment options and uses of information sources; their understandings of medical facts, of technologies applied to perinatal care and of prognosis; their views of life and living with handicaps; and information and communication needs of parents. Data related to parental needs during infants' NICU hospitalisation will be discussed by exploring the entire content of each interview.

Content analysis

Thematic content analysis (24) was performed using the software NVivo11 (QSR International, USA, 2015). A triangulation strategy was used to guarantee the rigour and quality of research - the first author led the analysis by identifying, sentence by sentence, parental needs experienced in NICU, and the last author collaborated on the development of coding framework. Firstly, quotations with similar meanings were inductively synthesized into categories, based on parents' perceptions about their needs in NICU. Secondly, using theoretical sensitivity in consultation with the existing literature about parental needs in intensive care (2,25), the categories were grouped into the following themes: support, assurance, comfort, proximity, and information and communication. The content of open-ended question of the NICU Family Needs Inventory was analysed using the same coding scheme. The most illustrative verbatim quotes were selected by two authors and revised by an English native speaker.

Ethical approval

The study was approved by the National Data Protection Commission and the Ethics Committees of all seven hospitals where data was collected. Written informed consent was obtained from all participants according to the World Association's Declaration of Helsinki.

Results

Needs evaluation through parent questionnaire

The majority of participants were less than 35 years of age, married or lived with a partner, had no previous children and lived at less than 15 minutes distance from NICU (Table 1). About three quarters were parents of singletons and approximately 40% experienced complications during pregnancy. The majority of parents did not have an extremely low birth weight or an extremely preterm delivery. Fathers were significantly less educated, were less likely to have lower white-collar occupations and considered themselves as belonging to a low/medium-low social class more frequently than mothers.

Participants attributed high importance to all NICU family needs subscales, increasing slightly from comfort (Median(IQR): 3.3(3.0-3.6)) to assurance (Median(IQR): 3.9(3.8-4.0)) needs. Mothers revealed a statistically significant tendency to value more information needs than fathers (Median(IQR): 3.8(3.6-3.9) vs. 3.7(3.5-3.9)).

Mothers and fathers without previous children rated support needs higher than those with previous children (Table 2). First-time fathers also revealed a significant tendency to value assurance and information needs, in comparison with fathers with previous children. Fathers of twins reported more frequently higher rates of comfort needs than fathers of singletons. Mothers aged 35 years or more attributed more importance to the needs of proximity, information and comfort than younger mothers. Less educated mothers were more likely to rate support and comfort needs as more important than more educated mothers. Support and assurance needs were also higher scored, respectively, by mothers living at a 15 minutes distance or more from NICU and those without pregnancy complications, in comparison with their counterparts.

The majority of needs considered as very important by ≥90% of parents were related to assurance and proximity (Table 3). Additional needs regarding support (n=5), comfort (n=3) and information (n=1) were reported by 3 mothers and 6 fathers who responded to the open-ended question.

Table 1. Participants' characteristics and NICU Family Needs Inventory scores, stratified by gender.

	Total n=207	Mothers n=118	Fathers n=89
Age (years), n (%)			
<35	145 (70.1)	84 (71.2)	61 (68.5)
≥35	62 (29.9)	34 (28.8)	28 (31.5)
Educational level (years), n (%)			
≤12	129 (62.3)	66 (55.9)*	63 (70.8)*
>12	78 (37.7)	52 (44.1)*	26 (29.2)*
Marital status, n (%)			
Single/Divorced/Widower	27 (13.0)	16 (13.6)	11 (12.4)
Married/living with a partner	180 (87.0)	102 (86.4)	78 (87.6)
Occupation, n (%)[a]			
Upper white collar	82 (42.7)	44 (41.1)*	38 (44.7)*
Lower white collar	56 (29.2)	40 (37.4)*	16 (18.8)*
Blue Collar	54 (28.1)	23 (21.5)*	31 (36.5)*
Subjective social class, n (%)			
Low/Medium-low	161 (78.9)	85 (73.9)*	76 (85.4)*
Medium-high/High	43 (21.1)	30 (26.1)*	13 (14.6)*
Distance from home to NICU (minutes), n (%)			
<15	106 (52.7)	58 (50.9)	48 (55.2)
≥15	95 (47.3)	56 (49.1)	39 (44.8)
Previous children, n (%)			
No	149 (72.0)	84 (71.2)	65 (73.0)
Yes	58 (28.0)	34 (28.8)	24 (27.0)
Multiple pregnancy, n (%)			
No	159 (76.8)	91 (77.1)	68 (76.4)
Yes	48 (23.2)	27 (22.9)	21 (23.6)
Pregnancy complications[b], n (%)			
No	118 (57.0)	69 (58.5)	49 (55.1)
Yes	89 (43.0)	49 (41.5)	40 (44.9)
Extremely low birth weight delivery[c], n (%)			
No	145 (70.0)	81 (68.6)	64 (71.9)
Yes	62 (30.0)	37 (31.4)	25 (28.1)
Extremely preterm delivery[d], n (%)			
No	162 (78.3)	93 (78.8)	69 (77.5)
Yes	45 (21.7)	25 (21.2)	20 (22.5)
NICU Family Needs Inventory Subscales[e], median (IQR)			
Assurance	3.9 (3.8-4.0)	3.9 (3.8-4.0)	3.8 (3.7-4.0)
Proximity	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.8 (3.6-3.9)
Information	3.7 (3.6-3.9)	3.8 (3.6-3.9)*	3.7 (3.5-3.9)*
Support	3.5 (3.1-3.8)	3.5 (3.1-3.8)	3.4 (3.2-3.7)
Comfort	3.3 (3.0-3.6)	3.3 (2.9-3.6)	3.4 (3.0-3.6)

Notes: IQR – Interquartile range; In each variable, the total may not add 207 parents, 118 mothers or 89 fathers due to missing values *p value <.05 for the comparison between mothers and fathers.

[a]Unemployed (n=36) and retired participants (n=1) were classified considering their previous main occupation. Students (n=2), housewives (n=4) and armed forces occupations (n=3) were excluded from this classification; [b]Infectious, placental, haemorrhagic and cardiovascular complications; [c]<1000g; [d]<28 gestational weeks; [e]Scale ranging from 1 - not important to 4 - very important.

Table 2. NICU Family Needs Inventory subscales scores stratified by gender, according to participants' characteristics.

	Mothers (n=118)					Fathers (n=89)				
	Assurance Median (IQR)	Proximity Median (IQR)	Information Median (IQR)	Support Median (IQR)	Comfort Median (IQR)	Assurance Median (IQR)	Proximity Median (IQR)	Information Median (IQR)	Support Median (IQR)	Comfort Median (IQR)
Age (years)										
<35	3.9 (3.8-4.0)	**3.8 (3.5-3.9)**	**3.7 (3.6-3.9)**	3.4 (3.0-3.8)	**3.3 (2.9-3.4)**	3.9 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.5 (3.2-3.8)	3.4 (3.0-3.6)
≥35	3.9 (3.8-4.0)	**3.9 (3.6-4.0)**	**3.9 (3.7-4.0)**	3.6 (3.1-3.8)	**3.6 (3.1-3.9)**	3.8 (3.5-3.9)	3.7 (3.6-3.9)	3.6 (3.5-3.9)	3.4 (3.1-3.7)	3.4 (3.0-3.6)
Educational level (years)										
≤12	3.9 (3.8-4.0)	3.8 (3.6-4.0)	3.8 (3.7-3.9)	**3.6 (3.3-3.8)**	**3.4 (3.1-3.7)**	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.7 (3.5-3.9)	3.4 (3.2-3.8)	3.4 (3.0-3.6)
>12	3.8 (3.8-4.0)	3.8 (3.5-3.9)	3.8 (3.6-3.9)	**3.2 (3.0-3.7)**	**3.1 (2.9-3.4)**	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.7 (3.5-3.8)	3.4 (3.1-3.7)	3.2 (2.9-3.6)
Marital status										
Single/Divorced/widower	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.5 (3.1-3.8)	3.3 (2.9-3.6)	3.8 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.5-3.8)	3.4 (3.2-3.7)	3.4 (3.0-3.7)
Married/living with a partner	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.6 (3.2-3.8)	3.1 (2.9-3.6)	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.5 (3.2-4.0)	3.3 (3.0-3.6)
Occupation[a]										
Upper white collar	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.3 (3.0-3.7)	3.2 (2.9-3.5)	3.9 (3.7-4.0)	3.7 (3.6-3.9)	3.7 (3.5-3.9)	3.5 (3.0-3.7)	3.3 (2.9-3.6)
Lower white collar	3.9 (3.8-4.0)	3.8 (3.5-3.9)	3.8 (3.6-4.0)	3.5 (3.1-3.8)	3.3 (2.9-3.7)	3.9 (3.7-4.0)	3.9 (3.6-3.9)	3.7 (3.6-3.9)	3.4 (3.3-3.9)	3.4 (3.1-3.5)
Blue Collar	3.9 (3.8-4.0)	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.5 (3.2-3.8)	3.4 (3.0-3.7)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.6 (3.5-3.8)	3.4 (3.2-3.7)	3.3 (3.0-3.6)
Subjective social class										
Low/Medium-low	3.9 (3.8-4.0)	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.5 (3.1-3.8)	3.3 (3.0-3.7)	3.9 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.5-3.9)	3.4 (3.2-3.8)	3.4 (3.0-3.6)
Medium-high/High	3.8 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.5 (3.0-3.8)	3.3 (2.9-3.4)	3.8 (3.7-3.9)	3.8 (3.6-3.8)	3.7 (3.6-3.9)	3.6 (3.0-3.7)	3.3 (2.9-3.5)
Distance home-NICU (min)										
<15	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	**3.4 (3.0-3.8)**	3.3 (3.0-3.6)	3.9 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.4 (3.1-3.8)	3.4 (3.0-3.7)
≥15	3.9 (3.8-4.0)	3.8 (3.6-4.0)	3.9 (3.7-4.0)	**3.6 (3.2-3.9)**	3.3 (2.9-3.7)	3.8 (3.5-4.0)	3.8 (3.5-3.9)	3.6 (3.5-3.8)	3.5 (3.2-3.7)	3.3 (3.0-3.5)
Previous children										
No	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.6-4.0)	**3.6 (3.1-3.8)**	3.3 (2.9-3.7)	**3.9 (3.8-4.0)**	3.8 (3.6-3.9)	**3.7 (3.6-3.9)**	**3.6 (3.3-3.8)**	3.4 (3.0-3.6)
Yes	3.8 (3.8-4.0)	3.8 (3.5-4.0)	3.7 (3.5-3.9)	**3.3 (3.0-3.5)**	3.3 (3.0-3.6)	**3.7 (3.5-3.9)**	3.7 (3.6-3.8)	**3.5 (3.4-3.8)**	**3.2 (2.9-3.5)**	3.2 (2.9-3.5)
Multiple pregnancy										
No	3.8 (3.8-4.0)	3.8 (3.5-3.9)	3.8 (3.6-3.9)	3.4 (3.0-3.8)	3.3 (2.9-3.6)	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.7 (3.5-3.9)	3.4 (3.1-3.7)	**3.3 (2.9-3.6)**
Yes	3.9 (3.9-4.0)	3.9 (3.6-4.0)	3.9 (3.7-4.0)	3.6 (3.2-3.8)	3.4 (3.0-3.7)	3.8 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.8)	3.7 (3.3-3.9)	**3.6 (3.1-3.7)**
Pregnancy complications[b]										
No	**3.9 (3.8-4.0)**	3.8 (3.6-4.0)	3.8 (3.7-4.0)	3.6 (3.1-3.8)	3.4 (2.9-3.7)	3.9 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.5-3.8)	3.4 (3.2-3.7)	3.4 (3.1-3.6)
Yes	**3.8 (3.7-3.9)**	3.8 (3.5-3.9)	3.8 (3.5-3.9)	3.4 (3.0-3.8)	3.3 (3.0-3.6)	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.6 (3.1-4.0)	3.4 (2.9-3.7)
ELBW delivery[c]										
No	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.7-4.0)	3.6 (3.1-3.8)	3.4 (2.9-3.7)	3.9 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.4 (3.2-3.8)	3.4 (3.0-3.7)
Yes	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.3 (3.1-3.8)	3.2 (2.9-3.4)	3.8 (3.6-3.9)	3.7 (3.5-3.9)	3.6 (3.5-3.8)	3.4 (3.2-3.6)	3.3 (3.0-3.4)
Extremely preterm delivery[d]										
No	3.9 (3.8-4.0)	3.8 (3.6-3.9)	3.8 (3.6-3.9)	3.5 (3.1-3.8)	3.3 (2.9-3.7)	3.9 (3.7-4.0)	3.8 (3.6-3.9)	3.7 (3.6-3.9)	3.5 (3.2-3.8)	3.4 (3.0-3.7)
Yes	3.9 (3.8-4.0)	3.8 (3.6-4.0)	3.8 (3.6-3.9)	3.4 (3.1-3.8)	3.3 (2.9-3.6)	3.8 (3.5-3.9)	3.6 (3.5-3.9)	3.6 (3.4-3.8)	3.4 (3.1-3.6)	3.1 (2.9-3.5)

Note: ELBW – Extremely low birth weight; IQR – Interquartile range; Scores are based on rating scale ranging from 1 - not important to 4 - very important; Bold types represent significant differences (p <0.05).

[a]Unemployed (n=36) and retired participants (n=1) were classified considering their previous main occupation. Students (n=2), housewives (n=4) and armed forces occupations (n=3) were excluded from this classification;[b]Infectious, placental, haemorrhagic and cardiovascular complications; [c]<1000g; [d]<28 gestational weeks.

Table 3. Summary of the most important needs reported by parents of very preterm infants.

	Quantitative data		Qualitative data
Family Needs	**Needs considered very important by ≥90% of participants**	**Needs identified in open-ended question**	**Needs added by parents during interviews**
Assurance (reflecting the parental need to feel confident, secure and hopeful about their infant's outcomes, which stems from their trust in the health care system)	• Parents: to have questions about their infants answered honestly; to know that their infant is being handled gently by healthcare providers; to be assured that the best care possible is being given to their infant; to feel that the hospital personnel care about their infant; to know the expected outcome for their infant • Mothers: to be told specific facts concerning their infant's progress; to feel there is hope		• To put babies in the "first place", above parents
Proximity (including the parental need to remain near the infant, both emotionally and physically)	• Parents: to see their infant frequently • Mothers: to receive information about their infant at least once a day; to hold their infant in their arms and against their skin as soon as they can • Fathers: to be called at home about important changes in their infant's condition		• Taking care of the infant like "normal" parents do (change diapers give bath, measure body temperature)
Information and communication (including parental need to obtain realistic information about their infant health and care and about the NICU environment)	• Parents: to know how the infant is being treated medically	• Father: "To have information about the meaning of all NICU machines' beeps"	• Practical information provided by other parents • Reinforcement of consistency and privacy • Be aware of the division of work between nurses and neonatologists; more interactions and a closest parents-neonatologists relationship • Information about all daily "insignificant" procedures
Support (encompassing references to resources, systems and structures needed by parents)	• Mothers: to be given directions about how they can provide care to their infant in the NICU	• Parents: "To have financial support on meals and parking" • Mother: "To have psychological support when parents lost a child" • Father: "To have religious support in decision-making process about blood transfusions" • Father: "To support mothers with special physical health conditions after delivery"	• Support from the Portuguese government: extend parental leave for mothers and fathers; help with social security bureaucracy; create a human milk bank • Support from extended family: inform relatives/friends; helping in daily activities • Regular support from psychologists and social workers
Comfort (including references to personal comforts that are important to parents)	• Parents: to see that the NICU staff provide comfort to their infant	• Father: "The noise in NICU do not help to the rest of babies and parents" • Father: "To have an entrance for health professionals away from baby's incubators" • Mother: "To have a private room for mothers with infants hospitalised in NICU away from the full-term mothers' regular ward"	• To enhancing privacy in NICU ward (barriers between the incubators, a single room per family near NICU, a mourning room, a setting for breastfeeding and kangaroo care) • Natural light and thermal insulation • A sitting room with comfortable furniture and eat and drink facilities

Semi-structured interviews with a subset of parents

All interviewed parents referred to support, and information and communication needs (Table 4). They mentioned the importance for extending parental leave for both mothers and fathers, covering the period of hospitalisation, to facilitate their presence in NICU (Table 4 – quote 1.1), as well as for having access to financial assistance for parking expenses (1.2), for both parents staying overnight near NICU (1.3), and for all recommended vaccines independently of infant's gestational age at birth (1.4). Interviewed parents also referred to the need for having a technician available in the hospital to help them with the social security bureaucracy (1.5). They felt that the medical equipment (1.6) and the ratio of nurses per baby (1.7) needed to be updated. Parents also requested a public human milk bank (1.8). The support of extended family, in particular the grandparents, was highlighted as an important need to spread information about the infant's development to other relatives and friends (1.9) and to help them dealing with daily activities like "cleaning the house and cooking" (Father, I26). Interviewees reported the need for regular support from psychologists and social workers beyond weekly meetings (1.10), which tended to be provided by nurses (1.11). The emotional support from other parents of infants hospitalised in NICU also emerged as an important need, mainly because the feeling of sameness facilitates mutual understanding and dialogue (1.12).

Interviewees highlighted the role of other parents on the provision of practical information about the expected infants' hospitalisation trajectory and emotional experience, both orally (2.1) or in-books (2.2). Parents mentioned the need for receiving coherent information provided by different health professionals about NICU rules and routines (2.3) and seemingly daily "insignificant" procedures such as changes in the localization of incubators (2.4), medical procedures (2.5) and discharge decision-making (2.6). They praised the provision of "the necessary information in a non-shocking way, in a way that do not over concern" them (2.7), but reported the need for reinforcing privacy when receiving information about infants' health status inside the neonatal ward (2.8). The awareness of the division of work between nurses and neonatologists was seen as an important information to improve communication: "Basically, in a daily basis we talked with the nurses (…) if it was a more serious thing [infection, heart problems], we talked to the doctor" (Father, I2). However, some interviewees claimed for more frequent interactions and a closer parent-neonatologist relationship (2.9).

Table 4. Illustrative quotations of the needs identified by the interviewed parents.

1. Support
1.1. Mother: I should have the right to have a bigger parental leave [100% instead of 65% of the salary], at least during the hospitalisation period. Mother stays there [NICU] for a lot of hours alone in a very difficult situation. At least during the hospitalisation in NICU, the father and the mother should have the right to stay both with a [full parental] leave (…). (I21)
1.2. Father: [Our home] It's too far [from NICU]. We have the car expenses, the tolls (…) Mother: The problem is that we have to pay the car parking [at the hospital] (…) Father: We can't [manage it]. (I6)
1.3. Father: For parents who lived far away [from NICU] there was a house for mothers to stay but (…) Mother: Perhaps allowing the father to stay as well. (I24)
1.4. Father: They [politicians] should include all the vaccines for all the babies, not only for those with less than 28/30 weeks, even because we have a lot of expenses with them [twins]. (I25)
1.5. Mother: Having a volunteer from the social security services in the hospital for explaining us [what kind of] papers we need to deliver [would be good]. (I10)
1.6. Father: Only the third ventilator [the staff tried] worked. (…) In an emergency, they had to try three ventilators and, in the middle of this, it could happen several things to the baby (…) the medical equipment is outdated. (I16)
1.7. Mother: There was one nurse for ten children (…) during the night. It's dangerous (…) During the day, [parents] help them [nurses] (…) but if something happens one nurse cannot do so much. (I6)
1.8. Mother: There are mothers with a lot of milk and instead of throw it away it could be for other children (…) It should exist a [public] human milk bank. (I7)
1.9. Father: They [grandparents] were giving the information [about twin infants] to the rest of the family and we were safeguarded for living only for them [infants]. (I25)
1.10. Father: The psychologist and the social worker only approach us [parents] for that [weekly] sessions and the parents need them in a daily basis, when the difficulties happen (…) when we are unhappy. Mother: We need them [psychologist and social worker] to be more present. (I24)
1.11. Mother: The Neonatology [unit] has a psychologist (…) but the nurses gave us a lot of support, every time we needed they gave us a lot of support. (I20)
1.12. Mother: The other mothers know what we are feeling because they are feeling the same (…) and we are more comfortable to talk with them [instead of a health professional]. (I6)
2. Information and communication
2.1. Mother: At the beginning (…) it was the other parents that explained to us what happened with their children, while the doctors were only saying that he [son] was stable. (I19)
2.2. Mother: When they [twin children] were in NICU I have read a book of testimonials of parents of preterm twins, people who know what I'm going through (…) and this was very important. (I10)
2.3. Father: We came [to NICU] in the day before [the birth] to understand what the unit was, how it worked etc… I know this doesn't happen with other parents (…) there are some professionals that value the family reception and others who don't. (I11)
2.4. Mother: One day, I arrived at NICU and I saw the incubator empty… I was in shock. I didn't make any question, I just started crying. When a nurse saw me [crying], she ran to told me that my daughter was moved to be closer to her twin. She should have careful and call me (…) or to the father. This could seem the most insignificant thing in the world but it's not, it's very important [for us]. (I7)
2.5. Father: When the doctor was going to do the ultrasounds nobody informed us. (I7)
2.6. Mother: [When] I came to the unit, the nurse told me the babies [twins] will go home (…) nothing predicted that because they didn't feed properly (…) suddenly they were going to be discharged. (I7)
2.7. Mother: People are well prepared to give us only the necessary information in a non-shocking way, in a way that do not over concern us (…) Giving us the information in a lightened way, it helps us. (I12)
2.8. Father: I understand that a NICU has to be an open space (…) but this fact implies that I am listening all the information about the baby besides mine. (…) The need to initiate antibiotic therapy is not the best thing in the world [for other parents hear]. (I22)
2.9. Mother: The nurses responsible for them [twin children] talked a lot with us [parents] (…) if we asked to talk to the doctor, he also come but he was more distant [from us]. I felt the need to the doctor be more [often] in contact with us. (I26)
3. Proximity

3.1. Mother: A normal mother changes the diaper (…) for us [mothers in NICU] to change a diaper [made us] feeling the best mother of the world … to measure the temperature (…). We [parents] gave her bath inside the incubator. (I1).
3.2. Mother: I couldn’t verbalized the word “son” during the first days (…) I only started to internalize this from the moment I could touch him, starting to feel him (…) the first time they [nurses] put him in skin-to-skin contact with me it was the moment I felt: this is real, he is mine. (I11)
3.3. Mother: We [parents] were there [NICU] just beside her [daughter], nothing more (…) and this is important (…). The fact that the hospital let the parents be there [NICU] 24h a day is great (…) it’s a tremendous luxury for parents. (I3)
4. Assurance
4.1. Mother: The respect for the babies is above all, above parents (…) Father: the most important [thing] is the baby, nothing more. Mother: We noticed there the babies are in first place, effectively, which give us assurance. (I3)
4.2. Mother: He [son] had two cerebral haemorrhages (…) Father: Yes, they [doctors] said to us that he had two little points in the ultrasound (…) [and then] in the morning they said to me that it was a little haemorrhage, without importance (…) after that we couldn’t trust [on doctors] because they didn’t tell us the truth (…) we lost the confidence on them. (I8)
4.3. Mother: I think that the [transfontanelar] ultrasounds performed by the medical students have to be done in the periods of parent’s absence (…) Father: It’s hard for parents watching the children being “butchered”. (I7)
5. Comfort
5.1. Father: I think that they [hospital administration] should create a physical barrier between the incubators. (…) If I want to cry there’s another parent beside the incubator of my infant looking at me or playing in the mobile phone. (…) It’s lack of privacy, it could have curtains. (I25)
5.2. Father: This hospital was 5 stars regarding privacy, she [mother] had a room only for her and in the other hospital the room is for 2 or 3 mothers. (I8)
5.3. Father: A mourning room for parents would be very pleasant. (I25)
5.4. Mother: The space for breast pumping and for doing the kangaroo care is very small (…) I was always being pushed because it was a passing area (…) and I would like a more private space. (I1)
5.5. Father: In other hospitals the light is artificial, here the light is good [natural] (…) but we can see that the windows, the air conditioning… Mother: Through those windows comes such a cold! Father: It’s old (…) it [physical environment] could be better. (I26)
5.6. Mother: I think it’s missing a room for parents. Father: Yes for, those who are there [NICU] all day, resting. The available room had only one chair, (…) without any furniture, only lockers. We need a coffee machine, a water machine (…) some chairs to talk to each other and get some rest. (I13)

Notes: I – Interview; NICU – Neonatal Intensive Care Unit.

Almost all interviewed parents underlined the importance for proximity needs (25/26 couples). They pointed out the need for taking care of the infant like “normal” parents do, by changing diapers, giving bath or measuring body temperature (3.1), while reinforcing the need for physical contact (e.g. touching and holding the baby, kangaroo care) to develop parental bonds (3.2), and for parental presence in the NICU without time restrictions (3.3).

More than a half of the interviewed parents (15/26 couples) revealed assurance needs. To “put babies first”, above parents (4.1), and to develop trustworthy parent-provider relationships were frequently mentioned. To feel confidence when leaving “the infants in doctors’ hands” (Mother, I10) was emphasised, which may be threatened when healthcare professionals didn’t tell parents “the truth” (4.2). Some interviewees talked about the need for having opportunity to decide whether they are willing or unwilling to stay beside the baby during painful procedures, such as the transfontanelar ultrasounds performed by medical residents (4.3).

A few interviewees (6/26 couples) mentioned comfort needs, calling for more privacy in NICU ward. Parents reported the need for having "physical barriers between the incubators" (5.1), a single room per family near NICU (5.2), a mourning room (5.3) and an appropriate setting for breastfeeding and for doing kangaroo care (5.4). They also mentioned the importance of natural light and thermal insulation in NICU ward (5.5), as well as a sitting room, with comfortable furniture and eat and drink facilities, to rest or to talk with other parents (5.6).

Discussion

Results from this mixed methods study can be useful for assessing and implementing quality family-centred and integrated healthcare services. Quantitative data suggest gender differences in factors associated with the importance attributed to parental needs: mothers valued more information needs than fathers and their overall scores were mainly influenced by age and level of education, while fathers' perception of their needs was mainly influenced by previous children. These findings contribute to a growing but still scarce literature addressing the specific factors associated with needs of parents of very preterm infants in NICU and support the development of further research on the degree of gender sensitivity of family needs measures. This study also showed that, despite gender differences, the assurance and proximity needs of parents apply across sociotechnical environments. All interviewees mentioned the need for instrumental support from the government for facilitating the presence of both mothers and fathers in NICU, and for regular emotional support from psychologists and social workers. These qualitative findings draw attention to the role of public policies in supporting or hindering parental involvement in NICU. Qualitative data also revealed additional needs related with information and comfort, alerting for the enhancement of privacy in the NICU ward and highlighting the role of other parents, nurses and neonatologists as complementary health mediators in the provision of detailed and coherent information about all NICU daily procedures. This knowledge helps in developing respectful and responsive family-friendly and gender-equality policies and healthcare, while challenging the items and the conceptual framework underlying the quantitative inventory.

Gender differences in the perception of parental needs in NICU have been previously reported (1,17,18). This study contributes to reveal how multiple femininities and masculinities intertwined with traditional gender roles, in the sense that persistence of mothers as primary caregivers (26) helps to explain why information regarding infants' health and NICU routines are more important for mothers than fathers, especially for the oldest ones. In addition, fathers without experience on parenting require being directly informed by staff, helping them to maintain control, to protect their family, and to participate in childcare (26). Healthcare professionals should be aware of the importance for clear, constant and reliable information adapted to infant's illness trajectory phase and parental cultural background (13,26).

In a context where parents of preterm infants tend to attribute higher scores to assurance and proximity needs worldwide (2,17), this study draws attention to the crucial role of formal support needs (e.g., parental leave policies, social security bureacreacy) and regular emotional counseling. This knowledge will contribute to promote policies that would assist families in their proximity needs (16), while reducing social inequalities and the stress created by financial hardship and dealing with bureaucracy in caring for a very preterm baby.

The need for reinforcing privacy during information provision as well as in NICU ward join the concern from recent studies revealing the importance of a consistent and continuous care (27) and underlining the confidentiality guarantee during handovers and ward rounds in NICU (28). Moreover, it challenges the idea that parents tend to neglect their comfort needs (1,17).

A limitation of this study is the time discrepancy between the two moments of data collection (during hospitalisation and 4 months after). Although it may shape differences between quantitative and qualitative results, very preterm infants stay hospitalised for long periods and parental experiences remain vivid by several months (29). Furthermore, the utility of the NICU Family Needs Inventory (2) for research and clinical purposes could be questioned due to its length and conceptual subjectivity underlying the placement of each item in its respective subscale (30).

In conclusion, this mixed methods study draws attention to family-friendly and gender-equality policies for supporting quality family-centred and integrated healthcare services in neonatology. Grounded on a consecutive and systematic recruitment of both mothers and fathers from all public level III NICU in North of Portugal during one year, this study raises awareness for the need of flexibility and sensitivity in developing conceptual frameworks and instruments to assess parental needs that take notice of socioeconomic position and reproductive trajectories of parents, as well as issues of privacy and regular emotional support in NICU.

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4.4. PAPER V: Quality of life of parents of very preterm infants 4 months after birth: A mixed methods study

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Quality of life of parents of very preterm infants 4 months after birth: a mixed methods study

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Abstract

Background: Knowledge about parental quality of life (QoL) is paramount to family-centred and integrated healthcare on prematurity, but evidence is limited. We aimed to explore mothers' and fathers' perspectives about their QoL 4 months after a very preterm childbirth.

Methods: This is a cross-sectional mixed methods study using a convergent design. Parents of very preterm infants were systematically recruited at all level III neonatal intensive care units in the Northern health region of Portugal for one year. Four months after childbirth, 61 mothers and 56 fathers filled-in the World Health Organization Quality of Life – BREF Inventory, and 26 couples were interviewed. Linear regression models were computed to assess the association between participants' characteristics and the QoL. Qualitative data were thematically analysed.

Results: A quantitative analysis revealed that the perception of QoL was not significantly different by gender. QoL scores increased slightly from the environment (Mean (SD): 72.1 (14.2)) to the psychological domains (Mean (SD): 78.7 (14.4)). All scores were influenced by psychological characteristics. Socioeconomic position influenced both parents' perceptions concerning the environment domain, and maternal physical and psychological QoL. Infant-related factors were associated with overall QoL among women and with the physical, psychological, social and environment domains among men. Qualitative findings indicated accommodation mechanisms that intertwine the focus on constraining factors (surveillance, sleep disturbances, non-supportive healthcare policies, hygienization) with facilitating factors (social support, accessibility/quality of healthcare, opportunities for developing parental skills). These processes were anchored in child-centredness and a framework that construct hierarchies of hope and expectations about infant's health and development.

Conclusions: To capture parental QoL using mixed methods raises awareness for developing intersectoral family-centred policies, integrated health services and focused-interventions to decrease the disempowering effects of surveillance and hygienization.

Keywords: Quality of life, Parents, Very preterm birth, Family-integrated care, Mixed methods

Background

Preterm birth is a major public health issue. Its complications constitute one of the leading causes of global deaths among children under 5 years of age [1], and preterm infants are at high risk of neonatal morbidity [2]. Globally, the average preterm birth rate in 2010 was estimated at 11.1%, corresponding to more than one in ten of all births [3], and about 1% were a very preterm birth, occurring before 32 gestational weeks [4]. Despite medical and technological advances, infants born very preterm remain at high risk of death and neurodevelopmental impairment, with studies revealing an average of crude in-hospital mortality rates of 14.2% in 10 European regions [5, 6]. A very preterm delivery and the ensuing child's hospitalization in a Neonatal Intensive Care Unit (NICU) is considered a disruptive and stressful life event, affecting parental QoL via multiple pathways [7, 8], in a context with wide differences between

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the support to family-friendly and gender-equality policies in Western and Nordic European countries [9].

The literature consistently shows that the quality of life (QoL) of parents of preterm infants may be compromised by sleep disturbances, fatigue, stress and psychiatric symptoms [10–14], while being protected by a stable marital union, support and information provided by medical staff, partner, extended family and other parents of preterm children [14, 15]. This knowledge is essential to develop family-centred and integrated healthcare services and policies on prematurity [16, 17], an approach with benefits for parents, children and their families [18, 19] as well as for healthcare staff and health services [20].

However, studies exploring the intertwining of constraining and protective factors in the perception of QoL are scarce and focused on patients with chronic conditions [21]. Furthermore, there is still limited evidence about the impact of a preterm delivery on parental QoL, in a context where methodological heterogeneity is observed regarding the operationalization of QoL and the use of units of analysis (mothers, parents, families and caregivers) [22]. Finally, the influence of fathers' characteristics and structural factors (e.g., parental leave policies) on parental QoL after a preterm delivery has not been sufficiently addressed in previous research [22].

Further studies are thus needed to explore both maternal and paternal QoL, in the analysis of the individual, familial and societal factors influencing QoL. Such in-depth parental perspectives are key, especially during the return-to-work period, which is a relevant moment in countries where few attempts are being made to support parental leave [23, 24]. A mixed methods approach would provide a more complete comprehension of the QoL questionnaires' scores, contributing to accurately capture the singular experience of parenting a very preterm infant [25] and the complexity of QoL assessment [26]. By integrating quantitative and qualitative data, this study aims to explore mothers' and fathers' perspectives about their own QoL, 4 months after a very preterm delivery.

Methods

This observational and cross-sectional mixed methods study used a convergent design aiming to merge quantitative and qualitative data into one overall interpretation, in which the quantitative results were validated or expanded with the qualitative data [27, 28]. This single-phase design (i.e. the quantitative and qualitative methods were implemented during the same timeframe and with equal weight) was chosen with the intention to best understand the QoL of parents of very preterm infants during the return-to-work period, ending up with well-substantiated conclusions about the factors that influence such phenomenon.

Between July 2013 and June 2014, all mothers and fathers of very preterm infants, admitted to all level III NICU located in the Northern Health Region of Portugal ($n = 7$), were consecutively and systematically invited to participate in the study by the healthcare team, 15 to 22 days after delivery. Parents who were present in the NICU during the hospitalisation period, who were able to speak and write in Portuguese, and those whose single or twin infants survived were considered eligible to participate in the study [29]. Among the 122 families invited, 96% agreed to participate in the evaluation at 4 months after delivery, the common return-to-work period in Portugal, in particular for mothers.

Clinical records were reviewed to retrieve data on pregnancy complications, multiple pregnancy, and infant's gestational age and birth weight. Extremely low birth weight was defined as birth weight bellow 1000 g and extremely premature infants were those with gestational age under 28 weeks [4, 30].

Quantitative study: Participants and data collection

Parents were contacted 4 months after delivery to confirm the availability to receive the questionnaires at home. Parents whose infants were still hospitalized ($n = 1$) or died ($n = 3$) were excluded from the study. Self-administered questionnaires to be completed individually, with prepaid return envelops, were sent by postal mail to 113 families. Among these, 67 mothers and 64 fathers completed and returned the questionnaires between November 2013 and November 2014 (Median months after childbirth (P25-P75): 4.3 (4.0–4.6)). After exclusion of the participants with > 20% of missing values on the QoL questionnaire, as recommended [31], 61 mothers and 56 fathers were included in the quantitative analysis.

Perceived QoL was assessed using the Portuguese version of the World Health Organization Quality of Life – BREF Inventory (WHOQOL-BREF) [32]. It is organized into a facet of *overall QoL* (general perception of QoL and health) and 4 domains: *physical* (pain and discomfort; energy and fatigue; sleep and rest; dependence on medication; mobility; activities of daily living; working capacity), *psychological* (positive and negative feelings; self-esteem; thinking, learning, memory and concentration; body image; spirituality, religion and personal beliefs), *social relationships* (personal relations; sexual activity; social support), and *environment* (financial resources; information and skills; recreation and leisure activities; home environment; accessibility and quality of health and social care; physical safety and security; physical environment; transport).

Data on sociodemographic characteristics were collected, as well as data regarding infants' length of stay in NICU and the presence of health problems.

Occupations were classified according to the Portuguese Classification of Occupations 2010 [33] and grouped in three categories: upper-white-collar, including executive civil servants, industrial directors and executives, professionals and scientists, middle management and technicians; lower-white-collar, including administrative and related workers, service and sales workers; and blue-collar, which includes farmers and skilled agricultural, fisheries workers, skilled workers, craftsmen and similar, machine operators and assembly workers, unskilled workers. Unemployed ($n = 15$) or retired participants ($n = 1$) were classified considering their previous main occupation.

Symptoms of anxiety, depression and parenting stress were assessed through Portuguese versions of The Beck Anxiety Inventory [34], the Beck Depression Inventory-II [35], and The Parenting Stress Index (PSI) [36], respectively.

Participants were georeferenced according to the home address, using the ArcGIS Online World Geocoding Service and Google Maps. Each participant was matched to the urbanity level [37] and the neighbourhood socioeconomic deprivation, assessed through The European Deprivation Index [38].

Statistical analysis

Missing values of the WHOQOL-BREF inventory were replaced by means of the remaining domain items, when ≤ 2 items were missing from the domains *physical, psychological* and *environment* and 1 item in the *social relationships* domain [31]. Regarding the BDI and BAI scores, participants with > 2 items missing were discarded from the current analysis; the remaining missing values were replaced by the mean value for each item [35]. Missing values in the PSI were substituted using the subscale items if no more than 5 items from total scale, 3 items from each domain and 1 item from each subscale were missing [39].

The analysis was performed using Stata 11.0 (College Station, TX, 2009). The chi-square test and the t-test or the Mann-Whitney-test were used as appropriate. Linear regression models, stratified by gender, were computed to assess the association between participants' characteristics and the QoL. Statistical significance was set at a value of $p < .05$.

Qualitative study: Participants and data collection

Semi-structured qualitative interviews were conducted with a sub-sample of 26 couples between November 2013 and April 2014. Participants were purposively sampled to include parents of infants with extremely (< 1000 g) and non-extremely (≥ 1000 g) low birth weight. A heterogeneity sampling was used for maximum variation of views and experiences, until reaching thematic saturation. Therefore, recruitment continued until no new themes emerged from the interview data [40].

Interviews were conducted at parents' home ($n = 19$), at the university department responsible for the study ($n = 6$) and in a private hospital room ($n = 1$). Interview duration ranged from 20 to 72 min (Mean: 39 min). All interviews were audio taped and transcribed verbatim. The interview guide covered the following areas: how parents deal with uncertainty and doubts and how they made their decisions concerning parental care, treatment options and uses of information sources; their views of the consent procedures; their understandings of medical facts, of technologies applied to perinatal care and of prognosis; their views of life and living with handicaps; information and communication needs of parents; and awareness of social and ethical issues in this area. Data related to parents' perceptions of their QoL will be discussed by exploring the entire content of each interview.

Content analysis

Thematic content analysis [41] was performed using the software NVivo 11 (QSR International, USA, 2015). A triangulation strategy was used to guarantee the rigour and quality of research - the first author identified, sentence by sentence, parents' perceptions about the factors influencing (positively and negatively) their QoL after a very preterm delivery, and the last author collaborated on the development of the coding framework. Firstly, quotations with similar meanings were synthesized into categories, both deductively, in accordance with the facets of the WHOQOL-100 inventory [31], and inductively for the remaining data. Secondly, the categories were grouped into the following analytical themes: the domains of the WHOQOL-BREF inventory (Physical, Psychological, Social relationships and Environment) [31] and "Accommodation mechanisms", corresponding to behavioural, cognitive, and emotional processes to accommodate a very preterm delivery [21]. The re-examination of qualitative data was performed when disagreements with quantitative results were found. The most illustrative verbatim quotes were selected by two authors and revised by an English native speaker.

Results

The characteristics of the parents who completed the questionnaire and their association with QoL are presented in Tables 1 and 2, respectively. The results are explored integrating quantitative and qualitative data, according to QoL domains.

Overall QoL and accommodation mechanisms

A quantitative analysis revealed that the perception of overall QoL was not significantly different by gender (Mean (SD): 74.6 (12.5) for mothers; 72.8 (12.4) for fathers). Higher levels of anxiety and depressive symptoms were negatively associated with the parental perception of overall QoL. Among mothers, having higher levels of

Table 1 Characterization of the participants who filled in the questionnaire, according to gender

	Total *n* = 117	Mothers *n* = 61	Fathers *n* = 56
Age < 35 years, n (%)	71 (62.8)	42 (68.9)	29 (55.8)
Educational level ≤ 12 years, n (%)	69 (60.5)	34 (55.7)	35 (66.0)
Married/living with a partner, n (%)	105 (92.1)	56 (91.8)	49 (92.5)
Occupation[a], n (%)			
Upper white collar	46 (42.2)	22 (37.9)*	24 (47.1)*
Lower white collar	32 (29.4)	25 (43.1)*	7 (13.7)*
Blue Collar	31 (28.4)	11 (19.0)*	20 (39.2)*
Low/Medium-low subjective social class, n (%)	87 (77.7)	43 (71.7)	44 (84.6)
Neighbourhood socioeconomic deprivation, n (%)			
T1 (Least deprived)	50 (42.7)	27 (44.3)	23 (41.1)
T2	38 (32.5)	20 (32.8)	18 (32.1)
T3 (Most deprived)	29 (24.8)	14 (23.0)	15 (26.8)
Urbanity Level, n (%)			
Predominantly Rural/Moderately Urban	15 (12.8)	8 (13.1)	7 (12.5)
Predominantly Urban	102 (87.2)	53 (86.9)	49 (87.5)
Parenting stress			
Total stress scale[b], Median (P25-P75)	216.5 (189.0–247.0)	220.0 (204.0–245.0)	209.0 (188.0–254.0)
Stressful life events scale[c], Median (P25-P75)	10.0 (4.0–15.0)	11.0 (4.0–19.0)	10.0 (4.0–15.0)
Anxiety[d], Median (P25-P75)	3.0 (1.0–7.0)	3.0 (1.0–7.7)	2.0 (1.0–5.0)
Depression[e], Median (P25-P75)	4.0 (2.0–8.0)	6.0 (3.0–9.0)*	3.5 (1.0–6.0)*
Previous children, n (%)	29 (26.1)	16 (26.2)	13 (26.0)
Multiple pregnancy, n (%)	23 (19.7)	12 (19.7)	11 (19.6)
Pregnancy complications[f], n (%)	51 (43.6)	27 (44.3)	24 (42.9)
Extremely low birth weight delivery[g], n (%)	33 (28.2)	18 (29.5)	15 (26.8)
Extremely preterm delivery[h], n (%)	24 (20.5)	13 (21.3)	11 (19.6)
NICU length of stay < 2 months, n (%)	71 (61.7)	37 (61.7)	34 (61.8)
Infants' health problems[i], n (%)	25 (21.4)	15 (24.6)	10 (17.9)
Quality of life (WHOQOL-BREF)[j]			
Overall, Mean (SD)	73.7 (12.4)	74.6 (12.5)	72.8 (12.4)
Physical domain, Mean (SD)	77.1 (12.6)	75.9 (12.2)	78.3 (13.1)
Psychological domain, Mean (SD)	78.7 (14.4)	77.2 (14.8)	80.4 (13.9)
Social relationships domain, Mean (SD)	75.1 (17.1)	75.8 (17.9)	74.3 (16.4)
Environment domain, Mean (SD)	72.1 (14.2)	72.9 (13.9)	71.3 (14.6)

[a] Students, housewives and armed forces occupations were excluded; [b] The total stress score is the sum of the scores in two domains: child's characteristics and parent's characteristics, with higher scores indicating higher levels of parental stress (range for the total scale: 104 to 517); [c] Stressful Life Events scale is composed by 24 different life events likely to cause stress (e.g.: unemployment, divorce, death of a relative), with higher values indicating more stress in life (range for the total scale: 0 to 114); [d] Higher values indicate higher levels of anxiety symptoms (range for the total scale: 0 to 63); [e] Higher values indicate higher levels of depressive symptoms (range for the total scale: 0 to 63); [f] Infectious, placental, haemorrhagic and cardiovascular complications; [g] < 1000 g; [h] < 28 gestational weeks; [i] Inguinal and umbilical hernias, metabolic disease, ovarian cysts, bronchial dysplasia, autoimmune disease, cardiac disease, congenital malformation; [j] Higher values represent better QoL (Range: 0–100)

Notes: In each variable, the total may not add 117 parents, 61 mothers or 56 fathers due to missing values; The proportions may not add 100 due to rounding; SD, Standard Deviation; *p value < .05 for the comparison between mothers and fathers

total stress, higher stress life scores, an extremely low birth weight delivery and an infant with health problems or hospitalized in NICU for 2 months or more, was associated with worst overall QoL.

Four main mechanisms to accommodate the delivery of a very preterm infant on their lives were mentioned by the interviewed couples. Firstly, being optimistic by choosing to *"be very practical"* and *"to think positive" (I26)*, despite being scared:

> *"Despite these little scares [cold, urinary tract infection and conjunctivitis], everything is going positively, it is going well." (I18)*

Table 2 Crude association between characteristics of participants and quality of life, according to gender

	Quality of life (WHOQOL-BREF)									
	Mothers (*n* = 61) Crude b (95%CI)					Fathers (*n* = 56) Crude b (95%CI)				
	Overall QoL	Physical	Psychological	Social relationships	Environment	Overall QoL	Physical	Psychological	Social relationships	Environment
Age, years (< 35 vs. ≥35)	− 0.6 (− 7.6; 6.4)	− 1.7 (− 8.5; 5.1)	0.6 (− 7.7; 8.8)	−3.3 (− 13.2; 6.7)	0.1 (− 7.7; 7.8)	1.5 (− 5.6; 8.7)	1.8 (− 5.4; 9.1)	− 0.9 (− 8.3; 6.5)	1.9 (− 7.2; 10.9)	−7.7 (− 16.0; 0.6)
Educational level, years (≤12 vs. > 12)	− 3.2 (− 9.7; 3.2)	−0.5 (− 6.8; 5.9)	−2.8 (− 10.5; 4.8)	−3.0 (− 12.2; 6.3)	**− 10.0** **(− 16.8; − 3.2)**	− 5.3 (− 12.6; 2.0)	− 4.1 (− 11.5; 3.3)	− 3.8 (− 11.4; 3.8)	6.1 (− 3.1; 15.2)	**− 11.5** **(− 19.7; − 3.3)**
Occupation[a]										
Lower white collar vs. Upper white collar	− 4.7 (− 12.2; 2.8)	− 6.7 (− 13.8; 0.4)	−6.2 (− 15.0; 2.7)	−8.7 (− 19.3; 1.9)	**−12.1** **(− 19.6; − 4.6)**	− 8.7 (− 19.4; 2.0)	−3.8 (− 15.0; 7.3)	−1.4 (− 12.7; 9.8)	−1.0 (− 14.8; 12.8)	−10.6 (− 22.7; 1.5)
Blue collar vs. Upper white collar	− 1.7 (− 11.1; 7.7)	2.0 (− 6.9; 10.9)	−1.8 (− 12.9; 9.3)	−6.8 (− 20.2; 6.6)	**−10.9** **(− 20.4; − 1.5)**	−7.2 (− 14.7; 0.4)	−6.2 (− 14.1; 1.6)	−5.2 (− 13.1; 2.8)	3.1 (− 6.7; 12.8)	**−14.1** **(− 22.6; − 5.6)**
Subjective Social Class (Low/Medium-low vs. Medium-High)	−4.0 (− 11.1; 3.2)	**− 9.2** **(− 15.9; − 2.6)**	**− 9.1** **(− 17.3; − 0.8)**	− 9.1 (− 19.2; 1.0)	**− 15.7** **(− 22.5; − 8.8)**	−1.0 (− 10.8; 8.8)	− 2.2 (− 12.2; 7.8)	−1.4 (− 11.6; 8.8)	9.0 (− 3.2; 21.2)	− 8.0 (− 19.5; 3.6)
Parenting stress										
Total stress scale	**− 0.1** **(− 0.2; − 0.1)**	− 0.1 (− 0.2; 0)	**−0.2** **(− 0.3; − 0.1)**	**−0.2** **(− 0.4; − 0.1)**	**−0.2** **(− 0.3; − 0.1)**	−0.1 (− 0.2; 0)	−0.1 (− 0.2; 0)	**−0.2** **(− 0.2; − 0.1)**	**−0.2** **(− 0.3; − 0.1)**	−0.1 (− 0.2; 0)
Stressful Life Events scale[b]	**− 0.3** **(− 0.6; − 0.1)**	−0.2 (− 0.4; 0.1)	−0.2 (− 0.5; 0.2)	−0.3 (− 0.7; 0.1)	**−0.4** **(− 0.7; − 0.1)**	−0.1 (− 0.5; 0.2)	−0.3 (− 0.6; 0.1)	−0.2 (− 0.6; 0.2)	−0.3 (− 0.8; 0.2)	−0.4 (− 0.8; 0)
Anxiety	**−1.1** **(− 1.6; − 0.6)**	**−1.3** **(− 1.8; − 0.9)**	**−1.3** **(− 1.9; − 0.7)**	**−1.4** **(− 2.1; − 0.6)**	**−1.1** **(− 1.7; − 0.5)**	**−1.1** **(− 1.7; − 0.6)**	**−0.9** **(− 1.5; − 0.3)**	**−1.2** **(− 1.8; − 0.6)**	**−1.6** **(− 2.4; − 0.9)**	**−1.1** **(− 1.8; − 0.5)**
Depression	**−1.2** **(− 1.8; − 0.7)**	**−1.6** **(− 2.1; − 1.2)**	**−2.1** **(− 2.6; − 1.6)**	**−2.0** **(− 2.8; − 1.3)**	**−1.4** **(− 2.0; − 0.8)**	**−1.3** **(− 1.9; − 0.6)**	**−1.5** **(− 2.2; − 0.9)**	**−2.2** **(− 2.8; − 1.6)**	**− 1.9** **(− 2.7; − 1.0)**	**−1.5** **(− 2.3; − 0.7)**
Previous children (Yes vs. No)	−4.7 (− 12.0; 2.5)	0.3 (− 6.9; 7.4)	− 3.8 (− 12.5; 4.8)	0.3 (− 10.2; 10.8)	−4.6 (− 12.7; 3.5)	−1.5 (− 9.8; 6.8)	0 (− 8.5; 8.5)	−1.8 (− 10.5; 6.9)	−0.2 (− 10.5; 10.1)	−0.4 (− 10.3; 9.6)
Pregnancy complications[c] (Yes vs. No)	− 3.4 (− 9.9; 3.0)	1.2 (− 5.1; 7.5)	−3.8 (− 11.4; 3.8)	−0.9 (− 10.2; 8.4)	−2.9 (− 10.1; 4.3)	−0.7 (− 7.4; 6.1)	0.2 (− 7.0; 7.3)	4.8 (− 2.7; 12.3)	0.7 (− 8.3; 9.6)	2.0 (− 6.0; 10.0)
Multiple pregnancy (Yes vs. No)	5.7 (−2.3; 13.7)	− 4.8 (− 12.7; 3.0)	4.0 (− 5.6; 13.5)	−0.2 (− 11.8; 11.4)	3.3 (− 5.7; 12.3)	4.2 (− 4.2; 12.6)	3.5 (− 5.3; 12.4)	0.3 (− 9.1; 9.8)	2.8 (−8.3; 13.9)	0.1 (− 9.9; 10.0)
Extremely low birth weight delivery[d] (Yes vs. No)	**−8.3** **(− 15.0; − 1.5)**	0.7 (− 6.2; 7.6)	−0.5 (− 8.9; 7.9)	2.8 (− 7.3; 12.9)	3.7 (− 4.1; 11.5)	− 2.6 (− 10.2; 4.9)	−6.5 (− 14.3; 1.3)	−4.1 (− 12.4; 4.3)	−8.1 (− 17.8; 1.7)	−2.3 (− 11.2; 6.6)
Extremely preterm delivery[e] (Yes vs. No)	−3.1 (− 11.0; 4.7)	−2.9 (− 10.6; 4.8)	−2.1 (− 11.4; 7.2)	−1.9 (− 13.1; 9.4)	−3.1 (− 11.9; 5.6)	− 4.3 (− 12.7; 4.1)	0.7 (−8.2; 9.6)	−1.7 (− 11.2; 7.7)	−8.5 (− 19.4; 2.4)	−2.4 (− 12.4; 7.5)
Length of stay, months (≥2 vs. < 2)	**− 7.3** **(− 13.7; − 0.8)**	− 2.9 (− 9.4; 3.7)	−5.2 (− 13.1: 2.7)	− 6.3 (− 15.8; 3.2)	−4.2 (− 11.6; 3.2)	−4.6 (− 11.4; 2.2)	**−9.9** **(− 16.8; − 3.0)**	**−9.3** **(− 16.6; − 1.9)**	**−12.0** **(− 20.7; − 3.4)**	− 6.0 (− 14.1; 2.1)
Infants' health problems[f] (Yes vs. No)	**−8.3** **(− 15.5; − 1.1)**	−5.3 (− 12.5; 1.9)	−6.2 (− 14.9; 2.5)	− 7.7 (− 18.2; 2.8)	−5.5 (− 13.7; 2.7)	−1.8 (− 10.6; 6.9)	−6.2 (− 15.3; 2.9)	− 4.1 (− 13.8; 5.7)	**−13.3** **(− 24.3; − 2.3)**	**−11.8** **(− 21.6; − 2.0)**
Urbanity Level	−8.5	−8.8	− 9.8	−10.5	−8.4	− 5.6	−0.9	− 6.5	−0.5	−10.0

Table 2 Crude association between characteristics of participants and quality of life, according to gender *(Continued)*

	Quality of life (WHOQOL-BREF)									
	Mothers (*n* = 61) Crude b (95%CI)					Fathers (*n* = 56) Crude b (95%CI)				
	Overall QoL	Physical	Psychological	Social relationships	Environment	Overall QoL	Physical	Psychological	Social relationships	Environment
(Predominantly Rural/Moderately Urban vs. Predominantly Urban)	(− 17.8; 0.8)	(− 17.8; 0.3)	(− 20.8; 1.2)	(− 23.9; 2.9)	(− 18.8; 2.1)	(− 15.6; 4.4)	(− 11.6; 9.8)	(− 17.7; 4.7)	(− 13.9; 12.9)	(− 21.7; 1.6)
Neighbourhood socioeconomic deprivation[g]										
T2 vs. T1	−1.1 (− 8.6; 6.4)	−3.9 (− 11.1; 3.3)	− 7.9 (− 16.6; 0.7)	−6.2 (− 16.8; 4.4)	− 5.9 (− 14.1; 2.3)	4.2 (− 3.7; 12.0)	− 1.8 (− 10.2; 6.6)	3.8 (− 5.0; 12.6)	9.8 (− 0.3; 19.8)	− 0.7 (− 10.1; 8.7)
T3 vs. T1	−2.2 (− 10.6; 6.1)	2.7 (− 5.3; 10.7)	− 4.8 (− 14.4; 4.8)	− 3.7 (− 15.5; 8.1)	−2.7 (− 11.9; 6.5)	4.9 (− 3.4; 13.2)	1.2 (− 7.6; 10.0)	3.8 (− 5.6; 13.1)	9.9 (− 0.8; 20.5)	0.5 (− 9.4; 10.5)

[a]Students, housewives and armed forces occupations were excluded; [b]Stressful Life Events scale is composed by 24 different life events likely to cause stress (e.g.: unemployment, divorce, death of a relative); [c]Infectious, placental, haemorrhagic and cardiovascular complications; [d] < 1000 g; [e] < 28 gestational weeks; [f]Inguinal and umbilical hernias, metabolic disease, ovarian cysts, bronchial dysplasia, autoimmune disease, cardiac disease, congenital malformation; [g]From tertile 1 (T1) (least deprived) to tertile 3 (T3) (most deprived)

Notes: 95% CI, 95% confidence interval; Bold type indicates statistically significant associations (*p* value < .05)

Secondly, reordering goals by giving priority to the infant and learning to devalue stressful *"little things"* while attributing more value *"to the really important things"* such as seeing the infant breathing autonomously:

> *"The little things that stress us on daily life (...) nowadays we devalued it, we attribute more value to the really important things (...) [like] seeing him [son] breathing for himself [without medical support] every day." (I2)*

Thirdly, using comparisons between their infants and those with severe health problems to highlight how they are *"lucky" (I2)* and should *"thank God" (I24)*. Lastly, reframing expectations about the current and future development of their infant helped parents to deal with the experience of parenting a very preterm infant:

> *"He [son] had some little problems (…) but it's nothing of concern in terms of development. (…) We can't expect that he, with 4 months, matches with a 4 months term baby." (I11)*

> *"In the future, it [the concern] will be knowing if she [daughter] will develop the speaking skill (…) the growth we already know that it will be slow." (I13)*

Physical QoL

Based on a quantitative analysis, physical QoL was slightly higher among fathers (Mean (SD): 78.3 (13.1) vs. 75.9 (12.2) for mothers). This perception was negatively associated with higher levels of anxiety and depressive symptoms among mothers and fathers. Physical QoL was lower among mothers from a lower subjective social class and among fathers of infants hospitalized in NICU for 2 months or more.

Interviewees only mentioned negative factors influencing their physical QoL. The main issues presented by the parents included sleep deprivation, nightmares and poor sleep quality, as well as unpredictability and lack of time to perform daily activities or organizing the house. Some interviewees considered the infant's dependence on medical substances and medical aids as *"a daily challenge" (I19)*, and reported self-dependence of medication to manage headaches connected to the burden of parenting very preterm infants (I25). Few couples emphasized the deterioration of working capacity by feeling *"lost [and] disorientated"* to supervise employees *(I17)*, as well as the discomfort experienced when pumping breast milk, seen as a *"little sacrifice"* for the child *(I11)*, and the tiredness provoked by the intensive full-time caring of a very preterm infant:

> *"It's like a 24 out of 24 hours job and then the tiredness is different. (…) Because she is preterm, [the routine is] even more intense." (I24)*

Psychological QoL

The highest quantitative score among the parental QoL domains was observed in the psychological domain among both mothers (Mean (SD): 77.2 (14.8)) and fathers (Mean (SD): 80.4 (13.9)). Lower levels of psychological QoL were associated with higher levels of parenting stress, anxiety and depression, for both mothers and fathers. This domain was also negatively associated with mother's lower subjective social class, and with having an infant hospitalized in NICU for 2 months or more among fathers.

Interviewees mentioned the surveillance as a major constraining factor to psychological QoL. Parents were aware of the burden caused by surveillance but revealed difficulties in overcoming their *"instinctive"* need to control all social interactions established with the baby and the environment, as well as their distrust on relatives and friends to take care of the infant:

> *"[When other people hold my son] I usually stay like "a security dog" (…) it's like an instinct." (I16)*

> *"The environment is always controlled. (…) The house has to be clean every day. (…) We have thermometers all over the house." (I2)*

> *"I can't leave my daughter (…) with anybody. (…) I don't know why." (I6)*

Participants justified such difficulties by expressing negative feelings that involve fears and uncertainties around the return to the hospital, the infant's death or suffering or the infant's future development. A few interviewees also invoked thinking difficulties, a *"completely loss of personal autonomy"* related to the need to live according to their infants *(I11)* and mixed emotions:

> *"It's a whirlwind of emotions, and it's a challenge dealing with all that things." (I24)*

> *"[Having a very preterm infant] means happiness, means torment, anxiety and joy." (I25)*

Some respondents neutralized the negative influence of a very preterm delivery on psychological QoL by focusing on positive feelings, such as *"joy"* and *"happiness"*, and assuring self-esteem based on self-confidence as *"strong"* women and *"very careful and responsible"* mothers. Additional strategies were related with enacting spirituality/religion and personal beliefs (e.g. considering that *"things happen because they have to happen, and we have to face them" (I1)*), as well as acquiring parental autonomy by learning how to administrate medical treatments at home:

> *Father: "We are more self-sufficient if we do the things [administrate injections] at home, so I have learnt to give the injection. We don't need to go out with him [son] to do this medical treatment."*
>
> *Mother: "Neither we are dependent of other people." (I24)*

Social relationships QoL

Mothers and fathers presented similar values of social QoL (Mean (SD): 75.8 (17.9); 74.3 (16.4), respectively). This domain was negatively associated with higher levels of parenting stress, anxiety and depressive symptoms among women and men. Fathers of infants hospitalized for 2 months or more and with health problems presented lower levels of social QoL.

In interviews, parents mentioned the benefits of pragmatic or emotional support provided by family, friends, healthcare providers or other parents of very preterm infants:

> *"We have my parents-in-law, and sometimes my parents, helping us to take care of him [son], for allowing us to do other things [washing the car, rest]." (I26)*
>
> *"Now they [friends] are [acting] with normality, they are more positive (...) They try to relax us and transmit us security." (I21)*
>
> *"If we [parents] don't know what to do we can call the NICU professionals of where he [son] was [hospitalized] (...) anytime." (I8)*
>
> *"Sometimes we [parents] call them [other NICU parents] and ask them how they dealt with baby's cramps. We talk to each other a lot of times." (I25)*

Different perspectives toward personal relationships were reported: some couples stated that the very preterm childbirth strengthen their marital relationship, while others complained about the lack of time *"for each other"*. Likewise, parents distinguished between supportive personal networks and those who criticize them:

> *"We feel a great understanding about our concerns with hygiene, I think we always felt they [family and friends] understand us and that they do everything to facilitate [our life]." (I12)*
>
> *"We know that (...) a lot of people and family members criticize us because we are excessively careful [with the infant]." (I3)*

Environment QoL

The lowest quantitative scores on parental QoL were observed in the environment domain (Mean (SD): 72.9 (13.9) among mothers; 71.3 (14.6) among fathers). They were negatively associated with lower levels of education, having blue-collar occupations and higher levels of anxiety and depressive symptoms for both parents. Among mothers, lower scores of environment QoL were also associated with having lower white-collar occupations, a low/medium-low subjective social class, higher levels of parenting stress and higher stress life scores. Fathers of infants with health problems scored worst on environment QoL.

Interviewees focused on the influence of the accessibility and quality of health and social care. Parents recognized government financial support for infant's healthcare, namely for hospitalization, medication and vaccination, and their satisfaction with medical services as enabling factors, but pointed the negative influence of non-supportive parental leave policies and family allowance, as well as lack of coverage of *"special"* milk and all vaccines that preterm babies need and the absence of a *"fast track"* for very preterm infants in the emergency room.

> *"Due to the infant prematurity, the parental leave should be extended, for both mother and father. (...) I would start working next month and she [daughter] needs special care at least for one year." (I20)*

Some participants also mentioned the negative influence of the hygienization of bodies and spaces. The concern with the sterilisation of hands and objects and the avoidance of touch and closeness in the relationships with the infant adversely affected their QoL:

> *Father: "The care with sterilization of hands (...) [and] for not kissing him [son] - perhaps if he was a normal baby there are things that we didn't going through."*
>
> *Mother: "If something drops to the floor, it goes immediately to the laundry." (I2)*
>
> *"At the entrance room, they [visits] have to put the mask on and to wash and sterilise the hands and they're only allowed to see the baby, nobody can touch her [daughter]." (I20)*

Other issues presented by the interviewees included constraints on their participation in recreation and leisure activities. They often referred to isolation and the absence of a *"social life"* as threatening their QoL. A few participants overcome these by taking advantage of opportunities to share *"enjoyable"* moments together, such as *"watching a movie or talking to each other" (I12)*. A few couples also reported different perspectives regarding the home environment and financial resources by combining their negative and positive influence on QoL in a hybrid way. The need to rearrange small home spaces due to their infant's medical needs, like *"a medical oxygen cylinder" (I19)*, to become aware of their family's inability to fulfil infant's needs due to financial constraints and lack of support in transportation *"for taking the infant to the clinical appointments" (I9)* contributed to deteriorate QoL, while the access to conditions for creating a *"calm environment"* in the household and to financial resources positively affected QoL.

Some participants highlighted how the opportunities for acquiring new information and skills improved their sense of competence and control at home. These opportunities occurred either during infant's hospitalisation in NICU through the *"intensive course"* provided by health professionals or outside NICU by being offered the opportunity to clarify doubts about the baby by the paediatrician:

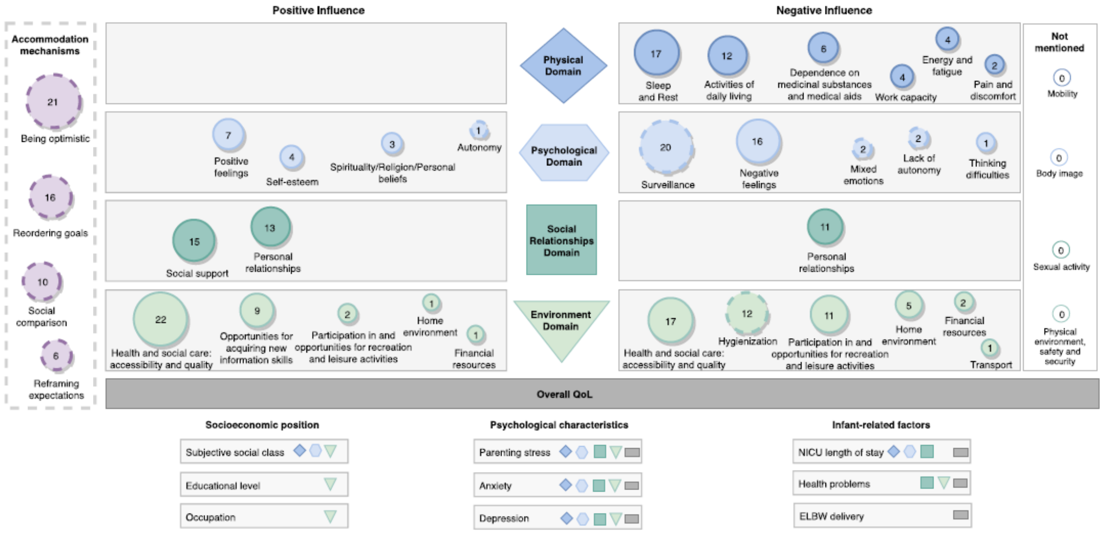
> *"We learned a lot [in NICU]. (...) It was there the father changed the first diaper, gave the first bath... He came home very prepared. (...) We used to say that it was an intensive course." (I18)*

> *"For us the most important thing is (...) having a person [health professional] to contact (...) anytime to clarify our doubts." (I5)*

Discussion

Quantitative data suggest that mothers and fathers of very preterm infants present similar values of QoL, increasing slightly from the environment to the psychological domain. Parenting stress, anxiety or depressive symptoms negatively influence both maternal and paternal QoL, while the impact of socioeconomic position and infant-related factors (NICU length of stay, health problems and extremely low birth weight delivery) varies according to gender and QoL domains. Qualitative findings highlight constraining factors related with surveillance, non-supportive healthcare policies and the need for hygienization, and protective factors as social support, accessibility and quality of healthcare, and opportunities for developing parental skills.

Participants' quantitative assessment of QoL is comparable to the scores observed in the Portuguese general population [32, 42, 43], reinforcing previous findings showing that there are no differences in QoL between parents of very low birth weight infants and the general population [44]. The negative association between depressive [11] or psychiatric symptoms [10] and QoL among mothers and caregivers of preterm infants has also been reported previously, as well as the influence of socioeconomic position [45]. Moreover, the stress-



Notes: Circles represent the frequency that each factor was mentioned in the interviews (full-line circles correspond to factors also assessed by the quantitative inventory; dashed-line circles correspond to factors only emerging in the qualitative interviews); ELBW - extremely low birth weight; NICU - neonatal intensive care unit; QoL - quality of life.

Fig. 1 Map of the factors influencing the QoL of parents of very preterm infants buffering effects of social support [46] and the positive impact of family-friendly and gender-equality policies [9, 47] on QoL are widely recognized.

This study adds to the literature the idea that similar quantitative scores of QoL might hide social inequalities and translate different meanings behind QoL. Figure 1 represents a comparison matrix in which a side-by-side joint display is used to converge the quantitative and qualitative data. First, each item assessed by the quantitative instrument does not acquire the same relative weight in parents' narratives. Second, there are facets assessed by the survey not mentioned by interviewees (mobility, body image, sexual activity, physical environment, safety and security). Third, parents mention several issues during the interviews that are not addressed by the questionnaire (such as the constant surveillance, hygienization of bodies and spaces, experience of mixed emotions, and lack of autonomy as negatively influencing their QoL). Bringing together the differing but complementing strengths of quantitative methods (e.g., trends and generalization) with those of qualitative methods (e.g. in-depth description and details) in a one-phase design might thus contribute to develop a specific quantitative tool to sensitively assess QoL of parents of very preterm infants, while helps to better understand their underlying factors.

When experiencing a very preterm childbirth, parents adjusted their expectations and changed their internal standards to accommodate such a catalyst event in their lives [21], as reported in studies with chronic illnesses patients [48]. The accommodation mechanisms observed in this study (being optimistic, reordering goals, social comparison and reframing expectations) are anchored in child-centredness, reflecting the incorporation of intensive parenting social norms and leading to the prioritization of child's health and well-being over parents' QoL [49], and in a pragmatic framework that construct hierarchies of hope [44] and expectations about infant's health status and development.

Couple interviews may have limited emergence of some facets assessed by the survey, in particular those related with body image and sexual activity. Interviewed parents may have felt uncomfortable acknowledging these issues in couple. In addition, the possibility of assuming as taken for granted facets as physical environment, safety and security cannot be excluded, as demonstrated by the quantitative rates. Further studies should explore the meanings attributed to each of these facets, discussing the implications for the assessment of the QoL.

These achievements reinforce the idea that the use of generic instruments may not be sensitive enough to accurately capture the specificities and idiosyncrasies of parents of very preterm infants [25, 50], overestimating their QoL. However, to acknowledge an individual holistic assessment that considers spirituality, religion and personal beliefs in QoL measurements is a step forward to improve the sensitivity of quantitative instruments, especially in health context [51–53]. Still, there is a need for further research on the development of a new quantitative tool specifically designed for being used to assess QoL among parents of very preterm infants.

The use of a convergent mixed methods design is a strength of this study, in which the inclusion of researchers who have quantitative and qualitative expertise addressed the effort to offer equal weight to two type of data. The sample size and the response rate could limit the power to detect small but potentially important differences, but they are quite similar to those observed in other studies with comparable populations and objectives [14, 54]. Moreover, there are no significant differences between participants who returned the questionnaire and those who did not regarding all the assessed variables except for marital status. Participants are more likely to be married or living with a partner (92.1% vs. 82.9% among non-participants, $p = .044$), which could cause some bias, since married people are more likely to score higher in the QoL questionnaires than people with other marital status [14, 55].

Conclusions

This study raises awareness for the need to capture the QoL of parents of very preterm infants using a mixed-methods approach for developing intersectoral family-centred public policies, integrated healthcare services on prematurity and focused-interventions to decrease the disempowering effects of surveillance and hygienization.

Abbreviations

NICU: Neonatal Intensive Care Unit; QoL: Quality of Life; WHOQOL-BREF: World Health Organization Quality of Life – BREF Inventory

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Availability of data and materials

The datasets generated and/or analysed during the current study are not publicly available due to a confidentiality agreement securing participants' privacy and anonymity but are available from the corresponding author on reasonable request.

Authors' contributions

MA, EA and SS collaborated in the acquisition, analysis and interpretation of the data. MA drafted the article. EA, MKI, AIR and SS reviewed the article critically for important intellectual content. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The study was approved by the National Data Protection Commission and the Ethics Committee of all hospitals where the data was collected. Informed consent was obtained from all individual participants included in the study.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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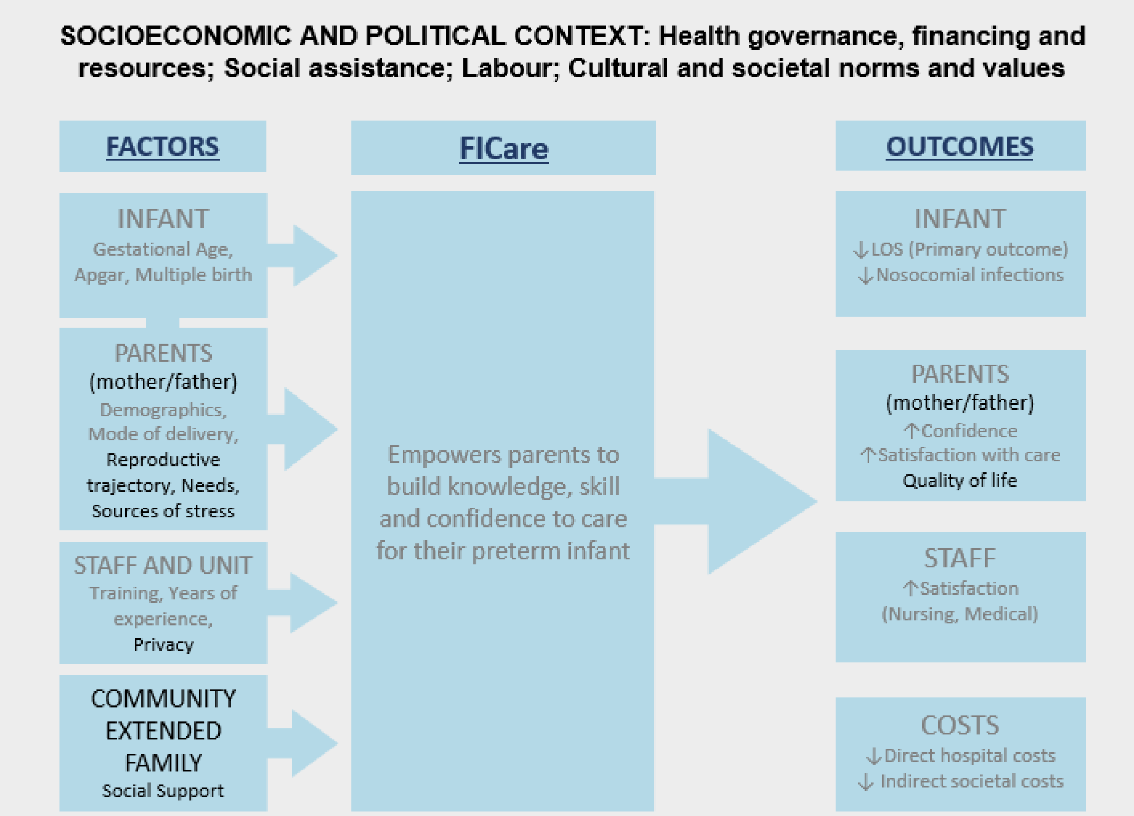
5. Discussion

5.1. What does this thesis add?

Achievements from this thesis provided evidence to sustain the development of policy and practice in family integrated neonatal services by analysing parental QoL following the birth of a premature infant. Taking a public health approach, it focuses on individual, familial, socioeconomic and political characteristics. This study adds to the conceptualization of FICare five key areas: analysis of parental QoL as an outcome; gendered sensitive assessment of parental needs and sources of stress in NICUs; inclusion of both mothers' and fathers' reproductive trajectories (i.e., multiple pregnancy and the existence of previous children) and privacy within staff and unit characteristics as influencing factors; take into account the community and extended family (e.g., social support); and introduction of the socioeconomic and political context (e.g., health governance, financing and resources; social assistance; labour; cultural and societal norms and values) (Figure 7).



Notes: FICare – Family Integrated Care; LOS – length of stay; Black type represents the issues added by the achievements from this thesis

Figure 7. Updated Family Integrated Care model (173)

The use of a mixed methods approach led to a wider understanding of the singular experience of parenting a very preterm infant inside and outside NICUs by allowing to cross different levels of analysis, identify issues missing in the available quantitative instruments, and explore, understand and disentangle the mechanisms behind inconsistencies and contradictions between quantitative and qualitative data. The comprehensive approach undertaken by this thesis renders its results applicable to various integrated neonatal care settings worldwide (236) and contributes for rethinking governance in neonatology (175, 183), by promoting the coordination of care both with and around the needs of infants, their families and communities to which they belong (237).

A major contribution of this thesis lies in the analysis of parental QoL as a main outcome of family integrated neonatal care. Traditionally, studies based on the FICare model are mainly concerned with infant-related outcomes such as infant weight gain, weight gain velocity, higher breastfeeding rates at hospital discharge, NICU mortality and major neonatal morbidities, safety, and resource use (including duration of oxygen therapy and hospital stay) as well as with parental stress and anxiety during NICU stay (71). However, QoL is a more meaningful outcome of integrated care models, as demonstrated by several studies with people with both physical and mental health comorbidities where more effective approaches were those that focus on QoL and enable people to live well with their conditions (237, 238). A great emphasis on the improvement of parental QoL inside and outside NICUs, involving strategies focused on the Raphael's concepts of "being" - emotional and physical well-being, "belonging" - interpersonal relationships, and "becoming" - personal development and self-determination (14), would contribute to diminish disparities in family health. Interventions focused on improving parental QoL typically involve non-professional care where members of the local community, such as parental associations, are a key resource in care delivery (237).

Social support, both instrumental and emotional, constituted a very important factor for sustaining family integrated neonatal care and parental QoL, especially for first-time parents. Support provided by extended family was highlighted as an important element to spread information about the infant's development to other relatives and friends and to help parents dealing with daily activities. Parents also reported the need for regular emotional support from psychologists and social workers beyond weekly parents' meetings, which tended to be supervised by nurses. The emotional and informational support from other parents of infants hospitalised in the NICU facilitated understanding and dialogue. These achievements, conjointly with previous studies showing the importance of the support system to parents' QoL and personal growth (239-241), reinforced the idea that the extended family and supportive networks (e.g., parental associations) should be included in the FICare model as factors associated with its successful implementation.

The literature tends to emphasise the microsystem-level parental needs worldwide, namely assurance, information and proximity (97, 99, 110), but this thesis raises awareness for the crucial role of macrosystem-related factors on parental QoL. Non-supportive public healthcare policies revealed to be a constraining factor for parental QoL inside and outside NICUs, reproducing the inequities regarding the support provided to family-friendly and gender-equality policies between Western and Nordic European countries (185). Parents appealed for extending parental leave for both mothers and fathers, covering the period of hospitalisation, to facilitate their presence in the NICU and their participation in care, as well as for having access to financial assistance for parking expenses, for both parents staying overnight near the NICU, and for all recommended vaccines independently of infant's gestational age at birth. According to Portuguese law, in case of having a hospitalised child, only one parent can benefit from a subsidy corresponding to 65% of his/her salary during the hospitalisation period, and usually the father returns to work 15 days after the childbirth (242). Under-debate currently is a law for a specific form of parental leave in the cases where parents have a premature childbirth or a newborn hospitalised (243), a long-time claimed right by these parents (181). Parents also referred to the need for having a technician available in the hospital to help them with the social security bureaucracy and requested a public human milk bank. This knowledge contributes to raise the attention for promoting policies that would increase parental QoL, assisting in their proximity needs, while reducing social inequalities and the burden and stress created by financial hardship and dealing with bureaucracy in caring for a very preterm infant (244).

Mothers and fathers of very preterm infants experience higher rates of psychological distress (symptoms of anxiety and depression) as well as slightly more stress than full-term parents (117, 245, 246). Despite the low levels of parenting stress, anxiety and depressive symptoms among participants of current study, the psychological factors had a negative influence on QoL, supporting previous literature that have stated a negative association between depressive (247, 248) and psychiatric symptoms (249) and QoL among parents of preterm infants (174). Thus, healthcare professionals need to be aware of the impact of parental mental health on QoL, especially during the first months after a very preterm childbirth, for identifying groups at risk that should constitute a privileged target for early interventions to promote QoL.

This thesis showed that the factors associated with the perception of parental QoL are different by gender, despite mothers and fathers presented similar values of QoL. Parenting stress, anxiety or depressive symptoms negatively influenced both maternal and paternal QoL while socioeconomic position and infant-related factors vary according to gender and QoL domains. Furthermore, gender was an important determinant of perceived sources of stress and needs, with mothers revealing higher levels of stress in all dimensions measured and valuing more informational needs than fathers. These differences contributed to reveal how multiples femininities and masculinities intertwined with traditional gender roles described in the literature when caring for a very preterm infant. The persistence of women as the primary caregivers (107) may explain why mothers of hospitalised infants are more often exposed to daily stressful circumstances than fathers (250, 251), needing more information regarding infant's health and NICU routines, especially the oldest ones. On the other hand, fathers tend to adopt the traditional role of breadwinner, being responsible to protect the whole family while simultaneously concerned with the child, the mother and the work/external environment (252), which make them perceive the hospitalisation as less stressful and needing less information from others than mothers. These insights reinforce the need for healthcare professionals to be aware of the importance for developing gender-sensitive strategies to reduce parenting stress, anxiety and depressive symptoms and to meet differential parental needs when aiming to improve both mothers' and fathers' QoL. In fact, the need for reinforcing privacy during information provision regarding infants' health status as well as in the NICU ward emerged as an important issue for parents of very preterm infants. Recent studies also underlined the importance of confidentiality guaranty during handovers and ward rounds in NICU (105, 253). These achievements challenge the idea that parents tend to neglect their comfort needs (99, 108), and highlight the importance of including privacy as a key issue in the guidelines for the design of NICU wards that consider the perspectives of their users (254).

Our results supported the non-association between infant-related factors and sources of stress and parental needs during the third week of NICU hospitalisation. However, infant's LOS and health problems assumed relevance for parental perceived QoL at 4 months after delivery, with longer hospitalisation periods and having infants with health problems being associated with lower levels of QoL among both mothers and fathers. These findings call the attention for healthcare services developing long-term follow-up programs assisting families beyond Neonatology Unit (93, 255), being particularly attentive to parents with these characteristics.

Accommodation mechanisms balanced the constraining and facilitating factors associated with parental QoL, although not assessed by the quantitative instruments. When experiencing a very preterm childbirth, parents adjusted their internal standards, values or conceptualization of QoL to accommodate such a catalyst event in their lives (31). Several accommodation mechanisms to cope with the very preterm childbirth were observed, such as being optimistic, reordering goals, social comparison and reframing expectations, which helped to maintain the levels of parental QoL similar to the general Portuguese population (229, 248). However, similar quantitative scores of QoL might hide social inequalities and translate different meanings behind QoL. On the one hand, there were issues assessed by the survey not mentioned by interviewees (mobility, body image, sexual activity, physical environment, safety and security); on the other hand, interviewees mentioned several issues not addressed by the questionnaire as negatively influencing their QoL, such as the constant surveillance, hygienization of bodies and spaces, experience of mixed emotions, and lack of autonomy. Thus, healthcare providers should be aware and incorporate those individual mechanisms in measurement and counselling programs specifically developed for families caring for a very preterm infant.

From a public health standpoint, this thesis raises issues that should be acknowledged on the co-production of intersectoral family-centred public policies, integrated neonatal care services and focused-interventions to promote QoL. An additional contribution of this study included the proposal of a short form of the *NICU Family Needs Inventory*, brief and easy to administrate, simple to score and to interpret. This instrument can contribute to increase parental participation in health research and to minimize the burden and the intrusion into parents' private lives (256), while allowing the identification and inclusion of family needs in counselling and clinical practice (40, 257).

5.2. Strengths and limitations

The findings of this thesis can be used in different integrated neonatal care settings, considering their comprehensive relationship with relevant literature in the field of governance in neonatology and FICare. A mixed methods approach maximized the differing but complementing strengths of quantitative methods (e.g., trends and generalization) with those of qualitative methods (e.g. in-depth description and details), contributing to a wider and sensitive assessment of needs and QoL of parents of very preterm infants, inside and outside NICUs, and their underlying factors.

The use of a representative sample of the Northern Portugal, with a very high response rate at baseline, reinforces the robustness of quantitative results. The recruitment of participants was carried out over an extended period of one year and parents were systematically invited to participate in all public level III NICUs from the Northern Health Region of Portugal. The fact that only 9.5% of participants had a non-Portuguese nationality have limited a discussion about the influence of migrant status and ethnicity on parental experiences in NICUs, an important variable which may be considered in the design of culturally sensitive family integrated neonatal care (82, 258). Replication of this study in other populations and health settings where more sociocultural diversity exists could help to sustain an international framework on FICare serving as a foundation for developing policies and practices to reduce inequalities and to promote parental QoL. The perspectives and experiences of NICUs' health staff and managers as well as parents of late preterm or sick babies hospitalized in NICU should also be taken into account in the dialogue about the development and implementation of family integrated neonatal care. In addition, other variables should be included as quality of marital relationship, parental health-related behaviours, and use of community-based developmental resources (e.g., early intervention programs).

The inclusion of fathers as well as mothers is a strength of this thesis. Previous studies about QoL among parents of preterm infants focused mainly on mothers (174), while research on health-related family issues has been analysing the couple as a unit, both in quantitative (259), qualitative (260) and mixed methods (210, 261) studies. Exploring both maternal and paternal perspectives and experiences enabled the identification of gender differences in the perception of sources of stress, needs and QoL, and allowed an examination of factors associated with these differences. In addition, this methodological feature offered the opportunity to discuss multiple femininities and masculinities associated with parenthood helping the design, development and implementation of gender sensitive healthcare practices and policies. This insight allowed a deeper understanding of the topics under analysis involving two different views and perspectives (211), but using the couple as the unit of analysis also involved some practical challenges and ethical dilemmas regarding voluntary participation, confidentiality and privacy (262-265). In addition, jointly couple interviews may have limited the emergence of some issues assessed by the quantitative questionnaires, as those related with body image and sexual activity, in particular among participants who may felt themselves uncomfortable acknowledging those issues in the presence of the partner. Finally, the possibility of non-independence between mothers and fathers cannot be excluded (266), claiming for future uses of dyadic analysis to explore the interdependency of maternal and paternal QoL perceptions (267, 268).

Although parental experiences during very preterm infants' hospitalisation in NICUs remain vivid for long periods (269), the possibility of differences between quantitative and qualitative results due to time discrepancy between the two phases of data collection (during the third week of hospitalisation and 4 months after childbirth) cannot be excluded. The use of different units of data collection and analysis in quantitative and qualitative data the individual and the couple, respectively, may have limited the comparison between the quantitative and qualitative results.

The response rate in the evaluation at 4 months after delivery and the consequent small sample size may have limited the power to detect small but potentially important differences in QoL perceptions. However, they are similar to those observed in other studies with comparable populations and objectives (270, 271). Moreover, there were no significant differences between participants who returned the questionnaire and those who did not regarding all the assessed variables except for marital status. Participants were more likely to be married or living with a partner, which could cause some bias, since married people are more likely to score higher in the QoL questionnaires than people with other marital status (271, 272). Moreover, due to underrepresentation of single families in this sample, further studies should consider to explore in detail the experience of this specific group of parents.

We did not aim to develop a new quantitative tool to assess QoL among parents of very preterm infants and future studies should explore it, to sensitively and accurately capture the specificities and idiosyncrasies of caring for a very preterm infant (36, 37), according to socioeconomic and political context in which families live in. To acknowledge an individual holistic assessment that considers spirituality, religion and personal beliefs in QoL measurements is a step forward to improve the sensitivity of quantitative instruments, especially in health context (22, 273, 274), but it still excludes issues related to accommodation mechanisms, surveillance, hygienization, mixed emotions and autonomy, which revealed to be important for these parents.

6. Conclusion

This thesis suggested that factors related with sociodemographic characteristics and reproductive trajectory influenced stress and needs of parents during the third week of very preterm infant's hospitalisation in a NICU, differently according to gender. Aspects related to infants and parental psychological characteristics were associated with parental QoL 4 months after the childbirth. Individual accommodation mechanisms and structural factors related with the socioeconomic and political context in which infants and families live in, especially regarding health governance, financing and resources, social assistance, labour as well as cultural and societal norms and values, influenced parental QoL throughout the experience of parenting a very preterm infant.

We identified the "change in parental role" as the major source of stress for both mothers and fathers during hospitalisation of a very preterm infant in a NICU. Mothers scored significantly higher in parental stress. Being older (≥30 years old) and having a multiple pregnancy were found to be significant predictors for decreased fathers' and mothers' stress, respectively. Based on our findings we recommend the development of sensitive instruments that take notice of gender and social support, and the implementation of interventions focused on reducing parental stress to diminish disparities in family health.

We have validated the NICU Family Needs Inventory for the Portuguese population and proposed a Short Form composed of 22 items and two dimensions, one focused on parents' needs and another on the infant's needs. Less educated and older parents more frequently attributed a significantly higher importance to parent-centred needs, while parents of multiples revealed a tendency to value infant-centred needs. The Short Form revealed to be a brief, simple and valid instrument, with a high degree of reliability, for being used in health research and practice.

Integrating quantitative and qualitative data, we explored the needs and QoL of mothers and fathers of very preterm infants. The importance attributed by parents to family needs increased slightly from comfort to assurance. Mothers valued more information needs than fathers. First-time fathers, as well as older and less educated mothers reported more needs than the younger and more educated ones. Despite gender differences, the assurance and proximity needs of parents apply across NICUs, as well as the need of instrumental support from the government; regular emotional support from psychologists and social workers; enhancement of privacy in the neonatology ward to assure family-centred information and comfort; and availability of other parents and health professionals as complementary health mediators in the provision of detailed and coherent information. This knowledge highlights the importance of family-friendly and gender-equality policies, and raises awareness of the need for flexibility and sensitivity in developing conceptual frameworks and instruments to assess parental needs that take notice of socioeconomic position and reproductive trajectories of parents, as well as issues of privacy and regular emotional support in NICUs.

Four months after delivery, parental QoL scores decreased slightly from the psychological to the environment domains. Parenting stress, anxiety or depressive symptoms negatively influenced both maternal and paternal QoL, while the impact of socioeconomic position and infant-related factors varied according to gender and QoL domains. Lower socioeconomic position negatively influenced both parents' perceptions concerning the environment domain, and maternal physical and psychological QoL. Infant-related factors (extremely low birth weight, hospitalisation in the NICU for 2 months or more, or infant's health problems) were negatively associated with overall QoL among women and with the physical, psychological, social and environment domains among men. Accommodation mechanisms activated by parents counterbalance constraining factors (surveillance, sleep disturbances, non-supportive healthcare policies, hygienization) with facilitating factors (social support, accessibility/quality of healthcare, opportunities for developing parental skills) of QoL. These processes were anchored in child-centredness and the construction of hierarchies of hope and expectations about infant's health and development. We reported that improvements on parental QoL depend on developing intersectoral family-centred policies, integrated health services and focused-interventions to decrease the disempowering effects of surveillance and hygienization.

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Appendices

Appendix 1: Study information sheet

Sou obrigado a participar?

Não. Caso decida não participar, esta decisão não terá qualquer influência em nenhum dos serviços de que usufrui. Mesmo depois de aceitar, poderá, em qualquer altura e sem justificação, desistir.

Quem é responsável pelo estudo?

O estudo é financiado pela **Fundação para a Ciência e a Tecnologia** e está a ser executado por investigadores da Faculdade de Medicina da Universidade do Porto (FMUP) e do Instituto de Saúde Pública da Universidade do Porto (ISPUP).

Como será usada a investigação?

Os resultados deste estudo serão divulgados junto dos profissionais que trabalham nestas áreas e aqueles que podem tomar decisões em relação à melhoria dos serviços prestados, nomeadamente políticos, médicos e os representantes da Secção de Neonatologia da Sociedade Portuguesa de Pediatria.

Obrigado por ter lido este folheto!

A sua participação será muito valiosa.

A aplicação do questionário apenas prosseguirá depois de colocadas todas as questões pelo participante e após assinatura do consentimento informado.

Ser-lhe-á dado este folheto informativo e uma cópia do consentimento informado.

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PAPÉIS PARENTAIS E CONHECIMENTO EM UNIDADES DE CUIDADOS INTENSIVOS NEONATAIS

(PACO)

Informação sobre o estudo

Caros pais e mães,

Está a ser desenvolvido um estudo com mães e pais de crianças nascidas entre as 22 e as 31 semanas de gestação e internadas nas Unidades de Cuidados Intensivos Neonatais (UCIN), sobre as suas experiências enquanto pais.

Gostaríamos de contar com a sua participação.

Antes de decidir se quer participar, é importante que saiba mais acerca deste estudo e do que lhe é pedido se aceitar participar.

Por favor leia atentamente este folheto informativo e coloque todas as perguntas que achar necessário.

Obrigado pelo tempo concedido à leitura desta informação!

Por que queremos falar consigo?

A finalidade deste estudo é dar voz às opiniões e conhecimento dos pais sobre os cuidados de saúde prestados nas UCIN. Em Portugal, este é o primeiro estudo nesta área.

Serão convidados a participar neste estudo as mães e pais de crianças muito pré-termo, internadas nas UCIN do Centro Hospitalar de S. João, Hospital Pedro Hispano, Hospital de S. Sebastião, Centro Hospitalar do Alto Ave, Centro Hospitalar do Porto - Unidade Maternidade de Júlio Dinis e Centro Hospitalar de Vila Nova de Gaia.

Entre outros aspetos, a informação recolhida será útil para:

- A saúde, na tentativa de enriquecer as boas práticas clínicas nas UCIN e a qualidade do acompanhamento disponibilizado aos pais, contribuindo para o desenho de cuidados intensivos neonatais mais eficientes;
- A ética médica, por permitir conhecer as necessidades de informação e comunicação dos pais;
- A igualdade social, para compreender as dificuldades e auxílios associados à prematuridade.

O que é que este estudo envolve?

Gostaríamos que respondesse a um questionário, que será aplicado por um(a) investigador(a), com uma duração máxima de 30 minutos. De uma forma geral, será inquirida/o sobre a sua experiência e conhecimento dos cuidados de saúde prestados nas UCIN.
Durante a aplicação do questionário, pode colocar todas as dúvidas e questões que deseje.
Como participante não terá que falar sobre assuntos que prefira não abordar.

A pesquisa é confidencial?

Sim. Toda a informação que partilhar connosco será vista somente pelos membros da equipa de investigação.
A informação será armazenada de forma segura.
Isto significa que, sempre que se utilizar as informações recolhidas, nunca será usado o seu verdadeiro nome.

Quais serão os benefícios da minha participação?

Será participante de um estudo inovador no nosso país que procura compreender as experiências e opiniões de pais de crianças muito pré-termo, contribuindo para melhorar os cuidados de saúde ao dar resposta às necessidades de mães, pais e crianças.

Appendix 2: Structured questionnaire developed by research team

Departamento de Epidemiologia Clínica,
Medicina Preditiva e Saúde Pública

QUESTIONÁRIO

Papéis parentais e conhecimento em Unidades de Cuidados Intensivos Neonatais

---

O Instituto de Saúde Pública da Universidade do Porto e a Faculdade de Medicina da Universidade do Porto estão a realizar um estudo sobre as experiências, opiniões e conhecimentos dos pais de crianças internadas em Unidades de Cuidados Intensivos Neonatais (UCIN) quanto aos cuidados de saúde prestados nestas unidades.

O seu contributo é extremamente importante para nós, mas completamente livre e voluntário. Toda a informação que nos fornecer será mantida sob anonimato, sendo portanto confidencial, e os dados recolhidos serão utilizados apenas para efeitos deste estudo. Desde já agradecemos a sua colaboração e o tempo que irá disponibilizar a responder a este inquérito.

Data de preenchimento: |___|___| / |___|___| / |___|___|

ID Inquiridor: |___|___|

Hora de início: |___|___|:|___|___|

Hora de fim: |___|___|:|___|___|

I. DADOS SOCIO-DEMOGRÁFICOS

Vou começar por lhe fazer algumas perguntas sobre a sua vida familiar e contexto profissional.

1. Qual a sua data de nascimento? |___|___| / |___|___| / |___|___|___|___|

2. De onde é natural?

   Portugal ☐ Outro país ☐

3. Onde reside atualmente?

   Distrito ________________________ Concelho ________________________

4. Qual é o seu estatuto marital?

Solteira	☐
Casada	☐
União de facto	☐
Viúva	☐
Divorciada	☐
Separada (casada, mas não vive com o cônjuge)	☐

4.1. Há quanto tempo estão casados/vivem em união de facto? |___|___| meses/anos

5. Qual o grau de escolaridade mais elevado que completou? NR ☐

Nenhum e não sabe ler, nem escrever	☐	Ensino secundário (12º ano)	☐
Nenhum, mas sabe ler e escrever	☐	Bacharelato	☐
1º ciclo do ensino básico (4º ano)	☐	Licenciatura	☐
2º ciclo do ensino básico (6º ano)	☐	Mestrado	☐
3º ciclo do ensino básico (9º ano)	☐	Doutoramento	☐

6. Qual a sua situação profissional atual? NR ☐

Empregada a tempo inteiro	☐
Empregada a tempo parcial	☐
Empregada menos que o tempo parcial (menos de 15 horas semanais)	☐
Trabalhadora familiar não remunerada	☐
Desempregada	☐
Estudante/ na escola/ em formação profissional	☐
Reformada e pré-reformada	☐
Doméstica/ocupa-se das tarefas do lar	☐
Outra situação	☐

7. Qual é/foi a sua profissão principal? (registar a profissão o mais detalhadamente possível) NR ☐

____

____

8. Qual o principal setor em que trabalha/trabalhava? NR ☐

Estado (Administração Pública central e local/entidades públicas autónomas)	☐
Empresa pública (ou empresas de capital maioritariamente público)	☐
Trabalhadora por conta de outrem no setor privado	☐
Trabalhadora por conta própria	☐
Doméstica – nunca trabalhou	☐
Não sabe	☐

9. É/foi supervisora/responsável pelo trabalho de outras pessoas?

Sim ☐ Não ☐ (passar à pergunta 11) NR ☐

10. Se é trabalhadora independente, quantos empregados tem?

Nenhum ☐ Menos de 10 ☐ 10 ou mais ☐ NR ☐

11. Vou agora fazer-lhe uma pergunta sobre um assunto que muita gente acha pouco simpático mas que é um dado útil para prever as condições de saúde das populações. Se quiser responder, diga-me, por favor, qual dos seguintes escalões corresponde ao rendimento mensal total líquido de todas as pessoas que vivem na sua casa?

A. ≤ 500 €	☐	E. 2001-2500 €	☐
B. 501-1000 €	☐	F. > 2500 €	☐
C. 1001-1500 €	☐	G. Não sabe	☐
D. 1501-2000 €	☐	H. Prefere não dizer	☐

12. Algumas pessoas consideram que a sociedade portuguesa está dividida em classes sociais. Das seguintes classes, em qual delas é que a senhora se incluiria?

A. Classe baixa	☐	E. Classe alta	☐
B. Classe média baixa	☐	F. Em nenhuma destas	☐
C. Classe média alta	☐	G. Prefere não dizer	☐

II. HISTÓRIA REPRODUTIVA

Vou agora colocar-lhe algumas questões que dizem respeito à sua história reprodutiva.

13. Quantas vezes já esteve grávida? |___|___| (se for a 1ª gestação passar à pergunta 15)

14. Qual o resultado obstétrico da(s) gravidez(es) anterior(es)?

	Gravidezes				
	1ª	**2ª**	**3ª**	**4ª**	**5ª**
Nado-vivo	☐	☐	☐	☐	☐
Feto morto > 22 semanas	☐	☐	☐	☐	☐
Abortamento espontâneo (“ovo branco”)	☐	☐	☐	☐	☐
Abortamento induzido	☐	☐	☐	☐	☐
Interrupção voluntária da gravidez	☐	☐	☐	☐	☐
Gravidez ectópica	☐	☐	☐	☐	☐
Doença do trofoblasto	☐	☐	☐	☐	☐

15. Alguma vez esteve a tentar engravidar durante mais de um ano sem conseguir?

Sim ☐ Não ☐ (passar à pergunta 16)

15.1. Se sim, quanto tempo? |__|__| (meses/anos)

16. Alguma vez consultou um médico por não conseguir engravidar?

Não	☐ (passar à pergunta 17)
Sim, em gravidezes anteriores	☐
Sim, nesta gravidez	☐
Sim, nesta gravidez e em anteriores	☐

16.1. Disseram-lhe qual a razão para não estar a conseguir engravidar?

Sim ☐ Não ☐ (passar à pergunta 17) Não sabe ☐ (passar à pergunta 17)

16.2. Que razão(ões) lhe apresentaram? NR ☐

Distúrbios hormonais que afetam a ovulação	☐
Obstrução das trompas	☐
Problemas do útero	☐
Endometriose	☐
Muco cervical que impede a passagem de espermatozóides	☐
Aborto de repetição	☐
Diminuição do número de espermatozóides	☐
Mobilidade reduzida dos espermatozóides	☐
Espermatozoides com configuração anormal	☐
Ausência de produção de espermatozóides	☐
Incapacidade de ejacular na vagina	☐
Outra. Qual? _____________________________________	☐

17. Alguma vez utilizou uma técnica de procriação medicamente assistida para tentar engravidar?

Sim ☐ Não ☐ (passar à pergunta 18) NR ☐

17.1. Quantos tratamentos já realizou?

1 ☐ 2 ☐ 3 ☐ >3 ☐ NR ☐

17.2. Em que gravidez(es)? NR ☐

Apenas em gravidez(es) anterior(es) ☐
Nesta gravidez ☐
Nesta gravidez e em anteriores ☐
Não se aplica ☐

17.3. Que técnica(s) usou? NR ☐

		Quantas vezes?
Inseminação artificial	☐	|___|___|
Fertilização *in vitro*	☐	|___|___|
Injecção intracitoplasmática de espermatozoide	☐	|___|___|
Transferência de embriões criopreservados	☐	|___|___|
Diagnóstico genético pré-implantação	☐	|___|___|

18. Quantos filhos tem? |___|___| (se não tem outro(s) filho(os) passar para a pergunta 20)

18.1. Dos seus filhos, quantos são:

Gémeos	|___|	Adotados	|___|
Concebidos por técnicas de PMA	|___|	De outra relação	|___|

19. Para além deste(s), algum dos seus filhos esteve internado numa Unidade de Cuidados Intensivos Neonatais?

Sim ☐ Não ☐ (passar à pergunta 20) NR ☐

19.1. Se sim, durante quanto tempo? |___|___| dias/meses

19.2. Qual foi o desfecho? NR ☐

III. ESTILOS DE VIDA

Vamos agora falar de alguns aspetos da sua vida, como o consumo de tabaco e a sua saúde.

20. Pense, por favor, em todos os aspetos da sua vida atual. Tudo somado, diria que se sente muito feliz, bastante feliz, pouco feliz ou nada feliz?

Muito feliz ☐
Bastante feliz ☐
Pouco feliz ☐
Nada feliz ☐
Não sabe ☐
Não responde ☐

21. Pense, por favor, em todos os aspetos da sua vida até aos 12 anos. Tudo somado, diria que se sentiu muito feliz, bastante feliz, pouco feliz ou nada feliz?

Muito feliz ☐
Bastante feliz ☐
Pouco feliz ☐
Nada feliz ☐
Não sabe ☐
Não responde ☐

22. Fuma ou alguma vez fumou?

Sim ☐ Não ☐ (passar à pergunta 24) NR ☐

23. Durante a gravidez alterou o consumo de tabaco?

Sim, parou de fumar ☐
Sim, reduziu o consumo ☐
Sim, aumentou o consumo ☐
Não, manteve o mesmo consumo que antes da gravidez ☐
Não, já tinha parado de fumar antes de engravidar ☐

24. Durante a gravidez,

24.1. familiares, amigos ou colegas de trabalho fumavam na sua presença?

Sim ☐ Não ☐ (passar à pergunta 24.2)

24.1.1. Com que frequência?

Diariamente ☐
5 a 6 vezes por semana ☐
2 a 4 vezes por semana ☐
1 vez por semana ☐
1 a 3 vezes por mês ☐
Menos de 1 vez por mês ☐

24.2. frequentou espaços onde era permitido fumar?

Sim ☐ Não ☐ (passar à pergunta 25)

24.2.1 Com que frequência?

Diariamente ☐
5 a 6 vezes por semana ☐
2 a 4 vezes por semana ☐
1 vez por semana ☐
1 a 3 vezes por mês ☐
Menos de 1 vez por mês ☐

25. Atualmente fuma?

Sim ☐ Não ☐ (passar à pergunta 25.2)

25.1. Quantos cigarros fuma, em média, por dia?

	Frequência
Cigarros	|___|___|,|___|
Outro. Qual?____________________________	|___|___|,|___|

25.2. Há quanto tempo deixou de fumar? |___|___|meses/anos

26. Bebe ou alguma vez bebeu bebidas alcoólicas, mesmo que apenas ocasionalmente?

Sim ☐ Não ☐ (passar à pergunta 29)

27. Durante a gravidez alterou o consumo de bebidas alcoólicas?

Sim, parou de beber	☐
Sim, reduziu o consumo	☐
Sim, aumentou o consumo	☐
Não, manteve o mesmo consumo que antes da gravidez	☐
Não, já tinha parado de beber antes de engravidar	☐

28. Atualmente consome bebidas alcoólicas?

Sim ☐ Não ☐ (passar à pergunta 29)

28.1. Com que frequência?

Diariamente	☐	1 vez por semana	☐
5 a 6 vezes por semana	☐	1 a 3 vezes por mês	☐
2 a 4 vezes por semana	☐	Menos de 1 vez por mês	☐

29. Alguma vez um(a) médico(a) lhe diagnosticou:

	Sim	**Não**	**NS**		**Sim**	**Não**	**NS**
Depressão	☐	☐	☐	Arritmia	☐	☐	☐
Diabetes	☐	☐	☐	Insuficiência cardíaca	☐	☐	☐
Dislipidemia	☐	☐	☐	Acidente vascular cerebral	☐	☐	☐
Hipertensão	☐	☐	☐	Cancro	☐	☐	☐
Enfarte agudo do miocárdio	☐	☐	☐	Outro(s). Qual(is)?_____________	☐	☐	☐

IV. ALEITAMENTO MATERNO

Vou agora colocar algumas questões sobre o aleitamento materno no contexto da UCIN.

30. O/a seu/sua filho/a foi alguma vez alimentado/a com leite materno?

Sim ☐ (passar à pergunta 31) Não ☐ (passar à pergunta 32) Não sei ☐ (passar à pergunta 33)

31. Se sim, foi necessário utilizar uma bomba extratora de leite?

Sim ☐ Não ☐

32. Se não, qual/quais o/s motivo/s?

Decisão materna	☐	Ausência de leite	☐
Decisão paterna	☐	Recusa do bebé	☐
Aconselhamento de familiares/amigos	☐	Outros__________________	☐
Recomendação médica	☐	Não responde	☐

33. Vou apresentar-lhe de seguida algumas situações que os pais podem considerar como facilitadores do aleitamento materno numa UCIN. Peço-lhe que indique, de entre estas, qual é, na sua opinião a que mais facilita o aleitamento materno na UCIN. E a 2ª? E a 3ª?

	1º Facilitador	2º Facilitador	3º Facilitador
Conhecer os benefícios da amamentação	☐	☐	☐
Contribuir para o crescimento e bem-estar da criança	☐	☐	☐
Poder tocar e estabelecer relações com a criança	☐	☐	☐
Aprender técnicas de extração e armazenamento do leite	☐	☐	☐
Incentivo dos profissionais de saúde	☐	☐	☐
Partilha de experiências com outros pais de crianças internadas	☐	☐	☐
Disponibilidade de materiais esterilizados para a extração do leite	☐	☐	☐
Envolvimento do pai no aleitamento materno	☐	☐	☐
Outro(s). Qual(is)?____	☐	☐	☐

34. Vou apresentar-lhe de seguida algumas situações que os pais podem considerar como dificuldades ao aleitamento materno numa UCIN. Peço-lhe que indique, de entre estas, qual é, na sua opinião a que mais dificulta o aleitamento materno na UCIN. E a 2ª? E a 3ª?

	1ª Dificuldade	2ª Dificuldade	3ª Dificuldade
Dificuldades no uso de bombas para extração do leite	☐	☐	☐
Preocupações quanto à produção e/ou extração de leite	☐	☐	☐
Informação inconsistente e/ou conselhos contraditórios	☐	☐	☐
Separação física das crianças	☐	☐	☐
Aspeto da criança	☐	☐	☐
Falta de apoio	☐	☐	☐
Outro(s). Qual(is)?______________________________________	☐	☐	☐

V. FONTES DE INFORMAÇÃO

Vou agora colocar algumas questões sobre as principais fontes que utilizou ou utiliza para obter informação relacionada com o internamento do/a seu/sua filho/a numa UCIN.

35. Vou apresentar-lhe de seguida algumas fontes de informação que os pais podem utilizar para obter informação relacionada com o internamento dos seus filhos numa UCIN. Peço-lhe que indique, de entre estas, qual é a principal fonte de informação que utiliza ou utilizou para obter informação relacionada com o internamento do/a seu/sua filho/a na UCIN. E a 2ª? E a 3ª?

	1ª Fonte	2ª Fonte	3ª Fonte
Médicos	☐	☐	☐
Enfermeiros	☐	☐	☐
Família, amigos e/ou colegas	☐	☐	☐
Pais de outras crianças internadas na UCIN	☐	☐	☐
Grupos de apoio	☐	☐	☐
Internet	☐	☐	☐
Folhetos e cartazes	☐	☐	☐
Outro(s). Qual(is)?____	☐	☐	☐
Nenhuma	☐	☐	☐

Appendix 3: Versão curta do Inventário de Necessidades da Família na Unidade de Cuidados Intensivos Neonatais (UCIN)

[Short form of the Neonatal Intensive Care Units (NICU) Family Needs Inventory]

Appendix A. Supplementary data

Versão curta do Inventário de Necessidades da Família na Unidade de Cuidados Intensivos Neonatais (UCIN)

Por favor assinale (X) quão IMPORTANTE é, para si, cada uma das seguintes necessidades.

	Nada importante (1)	Pouco importante (2)	Importante (3)	Muito importante (4)	Não se aplica (5)
1 – Poder entrar na unidade em qualquer altura.	________	________	________	________	_____
2 – Falar com o médico responsável pelo meu bebé todos os dias.	________	________	________	________	_____
3 – Ter uma sala de espera na unidade.	________	________	________	________	_____
4 – Saber quem são os profissionais que me podem dar informações sobre a saúde e bem-estar do meu bebé.	________	________	________	________	_____
5 – Ter um profissional específico a quem telefonar no hospital quando não posso visitar o meu bebé.	________	________	________	________	_____
6 – Estar disponível um grupo de apoio constituído por outras famílias.	________	________	________	________	_____
7 – Ter aulas sobre bebés prematuros e as suas necessidades de cuidados especiais.	________	________	________	________	_____
8 – Ter outra pessoa comigo quando visito a UCIN.	________	________	________	________	_____
9 – Saber exatamente o que está a ser feito pelo meu bebé.	________	________	________	________	_____
10 – Ter móveis confortáveis na sala de espera.	________	________	________	________	_____
11 – Receber a visita de um padre, pastor ou outra pessoa da minha comunidade religiosa.	________	________	________	________	_____
12 – Ter um telefone perto da sala de espera.	________	________	________	________	_____
13 – Sentir que sou aceite pelos profissionais do hospital.	________	________	________	________	_____
14 – Darem-me informação acerca de pessoas que possam ajudar a lidar					

com problemas relacionados com a minha situação.	________	________	________	________	_____
15 – Falar com o/a mesmo/a enfermeiro/a na maioria das vezes.	________	________	________	________	_____
16 – Falar acerca da possibilidade do meu bebé morrer.	________	________	________	________	_____
17 – Ter cadeiras confortáveis ao lado do berço ou incubadora do meu bebé.	________	________	________	________	_____
18 – Darem-me material para ler sobre a situação médica do meu bebé.	________	________	________	________	_____
19 – Permitirem-me ajudar nos cuidados físicos prestados ao meu bebé.	________	________	________	________	_____
20 – Ter um espaço para dormir perto da UCIN.	________	________	________	________	_____
21 – Saber que o meu bebé está a receber medicação para a dor.	________	________	________	________	_____
22 – Ter um espaço para estar sozinho(a) enquanto estou no hospital.	________	________	________	________	_____

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