**Elevated health-care use at five years of age in children born very preterm in a European cohort**

**Authors:** Anna-Veera Seppänen, MPH, PhD1, Elizabeth S Draper, PhD2, Stavros Petrou, PhD3, Henrique Barros, MD, PhD4, Adrien M Aubert, MSc1, Lazaros Andronis, PhD5, Sung Wook Kim, PhD3, Rolf F Maier, MD6, Pernille Pedersen, MD, PhD7, Janusz Gadzinowski, MD, PhD8, Jo Lebeer, MD, PhD9, Ulrika Ådén, MD, PhD10,11, Liis Toome, MD, PhD12,13, Arno van Heijst, MD, PhD14, Marina Cuttini, MD, PhD15 and Jennifer Zeitlin, MA, DSc1, on behalf of the SHIPS Research group\*

**Affiliations**

1. Université de Paris, CRESS, Obstetrical Perinatal and Pediatric Epidemiology Research Team, EPOPé, INSERM, INRAE, F-75004 Paris, France
2. Department of Health Sciences, University of Leicester, Leicester, United Kingdom
3. Nuffield Department of Primary Care Health Sciences, University of Oxford, Oxford, United Kingdom
4. EPIUnit-Instituto de Saúde Pública da Universidade do Porto, Porto, Portugal
5. Division of Clinical Trials, Warwick Medical School, University of Warwick, Coventry, UK
6. Children’s Hospital, University Hospital, Philipps University Marburg, Marburg, Germany
7. Department of Neonatology, Hvidovre Hospital, Hvidovre, Denmark
8. Department of Neonatology, Poznan University of Medical Sciences, Poznan, Poland
9. Department of Family Medicine & Population Health (FAMPOP), Disability Studies, Faculty of Medicine & Health Sciences, University of Antwerp, Antwerp, Belgium
10. Department of Women’s and Children’s Health, Karolinska Institutet, Stockholm, Sweden
11. Department of Neonatal Medicine, Karolinska University Hospital, Stockholm, Sweden
12. Department of Neonatal and Infant Medicine, Tallinn Children's Hospital, Tallinn, Estonia
13. Department of Pediatrics, University of Tartu, Tartu, Estonia
14. Department of Neonatology, Radboud University Medical Center, Nijmegen, the Netherlands
15. Clinical Care and Management Innovation Research Area, Bambino Gesù Children’s Hospital, IRCCS, Rome, Italy

**\***A complete list of study group members appears in the acknowledgements

**Address correspondence to:**

Anna-Veera Seppänen

Université de Paris, CRESS, Obstetrical Perinatal and Pediatric Epidemiology Research Team (EPOPé), INSERM, INRAE, F-75004 Paris, France

Maternité de Port-Royal, 53 Avenue de l’Observatoire, 75014, Paris, France  
E-mail: [anna-veera.seppanen@inserm.fr](mailto:anna-veera.seppanen@inserm.fr), tel. +33 (0)1 4234 5583

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**Abbreviations**

BPD Bronchopulmonary dysplasia

cPVL Cystic periventricular leukomalacia

ER Emergency room

GA Gestational age

GP General Practitioner

IQR Interquartile range

ISCED International Standard Classification of Education

IVH Intraventricular hemorrhage

NEC Necrotizing enterocolitis

ROP Retinopathy of prematurity

RR Risk ratio

SD Standard deviation

SGA Small for gestational age

VPT Very preterm

**Abstract**

**Objectives**

To describe parent-reported health-care service use at five years of age and investigate whether perinatal and social factors and routine follow-up are associated with elevated service use in children born very preterm. Few studies assess health service use beyond early childhood.

**Study design**

We used data from an area-based cohort of births <32 weeks’ gestation from 11 European countries, collected from birth records and parental questionnaires at five years of age. Using the published literature, we defined elevated outpatient/inpatient care (≥4 sick-visits to general practitioners, pediatricians or nurses, ≥3 emergency room visits, or ≥1 overnight hospitalization) and specialist care (≥2 different specialists or ≥3 visits). We defined level of routine follow-up coverage at five years by country, based on the rates of follow-up service use for children born very preterm.

**Results**

43% of children received elevated outpatient/inpatient care and 48% elevated specialist care during the past year. Perinatal factors were associated with outpatient/inpatient and specialist care but were more pronounced for specialist services. Associations with intermediate parental educational level and unemployment were stronger for outpatient/inpatient services. Living in a country with higher follow-up coverage was associated with lower use of outpatient/inpatient services.

**Conclusions**

Children born very preterm had high health-care service use at five years of age, with different patterns for outpatient/inpatient and specialist care by perinatal and social factors. Longer routine follow-up may improve care coordination and help avoid undesirable health service use.

**Introduction**

Very preterm (VPT) birth (<32 weeks’ gestation) can have heterogeneous and often multiple long-term consequences that reach beyond the neonatal intensive care hospitalization (1-5). Studies have shown that children born VPT require more health care in childhood (1, 6) and have higher health care-related costs compared to children born at or near term (7). However, research has mainly focused on hospitalizations and health service use in the first two years of life (8). A more comprehensive assessment of health service use in this population is important for identifying the range of services required, ensuring adequate resources and evaluating the accessibility, equity and quality of care.

Routine follow-up for children born VPT constitutes a key element of post-discharge care, aiming to identify health and developmental problems early, initiate early interventions and timely care and coordinate care provision. Efficient care coordination can help avoid preventable, or undesirable care, such as emergency room (ER) visits (14) and excess hospitalizations (14, 15). Early interventions have shown health and developmental benefits (16, 17), particularly in children from socially disadvantaged families, who have higher risks of adverse outcomes (5, 18-20). Thus, follow-up has the potential to improve care and mitigate the negative impact of social circumstances on health and development. However, follow-up (13, 21, 22), early intervention (23) and specialist (24) service use may be lower in children with social risk factors, whereas social risk factors have been associated with a higher risk of hospitalizations (25-27) and ER consultations (25, 28). Furthermore, European countries face challenges managing care for the children most reliant on health services manifested by a lack of post-discharge care coordination and policies for children with complex care needs (29), which could further compromise care provision and quality for those who need it the most (30). Nevertheless, few studies have addressed the role of routine follow-up in the equitable use of post-discharge health-care services.

We aimed to provide a comprehensive description of health service use among children born VPT at five years of age in 19 regions in 11 European countries, identify children with elevated health service use and assess perinatal and social factors associated with elevated service use. Given differences in routine follow-up policies for children born VPT at five years of age in participating countries (13), we also sought to determine whether living in a country with higher long-term routine follow-up coverage, based on observed rates of follow-up service use, affected these associations.

**Methods**

*Data source and study population*

The data were collected as part of the Screening to improve Health In very Preterm infantS (SHIPS) project, a five-year follow-up of the population-based Effective Perinatal Intensive care in Europe (EPICE) cohort of VPT births in 19 regions in 11 European countries (31): Belgium (Flanders); Denmark (Eastern Region); Estonia (entire country); France (Burgundy, Ile-de-France and the Northern region); Germany (Hesse and Saarland); Italy (Emilia-Romagna, Lazio and Marche); the Netherlands (Central and Eastern region), Poland (Wielkopolska); Portugal (Lisbon and Northern region); Sweden (greater Stockholm) and the United Kingdom (East Midlands, Northern, and Yorkshire & the Humber regions).

The cohort included all infants born between 22+0 and 31+6 weeks’ gestation in all maternity units in the participating regions over 12 months or six months (France only) between April 2011 and September 2012, according to local protocols. Out of 7,900 live births, 6,792 infants were discharged home alive and 6,759 were alive and invited to the follow-up at five years, of whom 3,635 (53.8%) participated (See Online Figure X). The five-year follow-up study took place over a twelve-month period, with starting dates between June 2016 and February 2017, reflecting country differences in the inclusion dates and the coordination of assessments with existing follow-up protocols.

*Data*

Pregnancy, perinatal and sociodemographic data were extracted from obstetric and neonatal records at baseline by trained study staff, using a standard, pretested questionnaire.

At five years of age, mothers, fathers or other caregivers responded to paper or online questionnaires about their child’s health and health service use and socioeconomic circumstances. Parents were asked how many times during the past year their child had seen each of the health-care providers suggested in a list of primary care providers (excluding routine check-ups and vaccination visits) and specialist physicians or health professionals (defined as those providing care not routinely provided by General Practitioners (GP) or pediatricians (8)) and selected to reflect specialist care most often provided to children born very preterm. Depending on the country, 15–17 care providers were suggested, based on options previously used in this cohort (8), reflecting local care practices relevant to the age group (see Table 1 [online]) for providers by country in national languages and English translations). Parents could report additional services as free-text responses. Free-text responses that corresponded to any of the services suggested in the questionnaire were recoded and included in the analyses. We did not include services reported only as free-text answers in the analyses, as few children used these services and the data was considered less reliable and comparable because it depended on parents recall without systematic prompts (see supplementary table X for a list of these services). The questionnaire included a separate question on overnight hospitalizations. A question on the child’s participation in routine follow-up for children born VPT was used to derive overall routine follow-up rates for the study countries, as reported elsewhere (13): “Does your child have routine check-ups for children who were born prematurely [*optional description of local service*]? (No, never had such check-ups, No, not anymore (please specify age at last check-up), Yes, still has check-ups (at neonatal unit where he or she was born, at other place or health care professional…))”

Perinatal factors included gestational age (GA) at birth, small for GA (SGA) status defined as birthweight between 3rd– 9th percentile (moderate SGA) and <3rd percentile (severe SGA) using intrauterine norms (32), bronchopulmonary dysplasia (BPD) defined as need for supplemental oxygen and/or assisted ventilation at 36 weeks’ postmenstrual age, a composite of other severe neonatal morbidities (retinopathy of prematurity (ROP) stages III–V, intraventricular hemorrhage (IVH) grades III–IV, cystic periventricular leukomalacia (cPVL), necrotizing enterocolitis (NEC) requiring surgery) (33), any congenital anomaly, child’s sex and multiple birth (singleton or one surviving multiple, twins, triplets or quadruplets).

Family sociodemographic factors included highest attained maternal educational level at five years based on the International Standard Classification of Education (ISCED) 2011 definition, categorized as: (I) lower: lower secondary (ISCED levels 0–2), (II) intermediate: upper or post-secondary, non-tertiary or short cycle tertiary (levels 3–5), and (III) higher: Bachelor degree or higher (levels 6–8) (18), parents’ employment status at five years (both parents/single parent employed, or at least one parent/single parent unemployed), maternal age at delivery, maternal country of birth (native, non-native European-born or born outside Europe), and parity at birth (nulliparous or multiparous).

*Health service use*

Because there is no consensus on what constitutes elevated health-care service use in pediatric populations born VPT, we adapted criteria from the published literature on complex care needs in the general pediatric population to define elevated service use, including definitions by Bramlett et al. (34), Kuo et al. (35), Patra and Greene (36) and Brenner et al. (30). Their definitions related to service use are described in Table 2. First, we used these criteria to describe the use of each health-care provider separately as (1) no, (2) occasional (1–3 sick-visits for GPs, pediatricians and nurses, 1–2 visits for ER services, once for overnight hospitalizations and 1–2 visits for specialists), and (3) frequent use (≥4 sick-visits for GPs, pediatricians and nurses, ≥3 for ER visits, ≥2 for overnight hospitalizations, ≥3 for specialist visits, and ≥2 for number of different specialists visited). These thresholds chosen for frequent use are higher compared to the literature, because children born VPT have higher health-care needs compared to the general pediatric population.

Using these adapted definitions, we further classified children as having elevated health service use for two types of services: I) outpatient/inpatient services (sick-visits to GPs, pediatricians, nurses or school nurses, ER visits and/or overnight hospitalizations) and II) specialist services. These definitions are described in Table 2.

We hypothesized that associations with social factors could be different for specialist services compared to sick-visits and acute care (ER visits and hospitalizations), based on the associations with social factors and use of specialist (24) and ER (25, 28) services and hospitalizations (25-27) in the published literature. We also hypothesized that routine follow-up could be differentially associated with sick-visits and acute care, in that this care may be prevented by improved care continuity and coordination (14, 15). In contrast, follow-up might increase use of specialist services as these target prematurity-related problems and are often an integral part of follow-up. We grouped sick-visits with ER and hospitalizations as acute care needs may be managed differently across countries; for instance non-urgent use of ER care that could have been provided by family doctors or pediatricians is common in some countries (37, 38) and pediatric hospitalization rates differ greatly (39).

*Analytical strategy*

We described the use of each service provider and type of service, including the percentage of children who had no, occasional or frequent visits as well as number of visits (mean, SD, median and IQR) and number of different specialists seen. We then investigated the proportion of children classified as having elevated outpatient/inpatient as well as specialist service use by country and in relation to the countries’ overall routine follow-up rates. We assessed crude rates of elevated outpatient/inpatient service use and specialist service use by perinatal risk and sociodemographic characteristics. We estimated unadjusted and adjusted risk ratios (RR) for both outcomes by perinatal risk and sociodemographic characteristics using multilevel generalized linear regression models taking into consideration clustering within mothers and countries, with a log link, Poisson distribution and robust standard errors (40). Adjustment variables included: GA at birth, SGA status, BPD, congenital anomaly, other severe neonatal morbidities, child’s sex, multiple birth, highest attained maternal educational level at five years, parents’ employment status at five years, maternal age at delivery, maternal country of birth, and parity at birth. We assessed associations with follow-up policy using a country-level variable on routine follow-up coverage in these models, based on parent-reported use of routine follow-up services at five years (at or above the mean, 27%: Portugal, Belgium, the Netherlands, France, Denmark and Sweden, versus below the mean: UK, Germany, Estonia, Italy and Poland). We performed a sensitivity analysis by repeating models for total number of care contacts for both types of services separately (i.e. higher number of visits defining elevated care), using multilevel negative binomial models, appropriate for count data where the variance is greater than the mean (41).

As the overall study response rate was 53.8% (range 29.3%–96.4% between countries) (31), we used inverse probability weights after multiple imputation (42), based on baseline characteristics, to give a higher weight to children with characteristics of non-responders in all analyses, as described elsewhere (43-45). The variables used for predicting inverse probability weights and for which missing data were imputed are reported in Supplementary Table Y. Adjusted models were also repeated without weights. STATA 14.2 was used for all analyses (Stata Corp., College Station, TX, USA).

*Ethics*

The work has been carried out in accordance with the Code of Ethics of the World Medical Association (the Helsinki Declaration). All study regions obtained ethical approval and parental written informed consent for follow-up according to national legislations. The study was also approved by the French Advisory Committee on Use of Health Data in Medical Research (CCTIRS) and the French National Commission for Data Protection and Liberties (CNIL).

**Results**

Compared to study participants, children whose parents did not respond to the five-year questionnaire were to a larger extent born a later GA to younger, foreign-born and multiparous mothers (see Online table 1).

In five year-old children born VPT, health-care visits (excluding routine visits and vaccinations) during the past year were frequent; 67.2% of the children had four or more health-care visits in total for a median of six contacts (IQR 3–15); 44.2% (median 3; IQR 1–6) had four or more outpatient/inpatient visits and 43.4% (median 2; IQR 0–8) had three or more visits to specialists listed in the questionnaire (Table 3). Eye specialists and ear/hearing specialists were the most consulted specialists for occasional (1–2) visits (32.6% [median 0, IQR 0–1] and 22.1% [median 0, IQR 0–1] respectively), and speech therapists (17.2% [median 0, IQR 0–0]) and physiotherapists (11.6% [median 0, IQR 0–0]) for frequent (three or more) visits. Whereas 28.8% had seen one specialist, 40.7% had consulted two or more different specialists [median 1, IQR 0–2]. Some parents reported other specialist care as free-text responses, including cardiologists or endocrinologists (<2% per provider, listed in supplementary table X). For outpatient/inpatient services, GPs and pediatricians were the most commonly used, regardless of number of visits, with over half of the children having at least one visit during the past year. One out of four children had one to two ER visits, and 5.4% had three or more ER visits (median 0; IQR 0–1); almost 10% were hospitalized overnight once over the past year, and 1.8% were hospitalized more than once (median 0; IQR 0–0).

Overall, 43.2% of the children had elevated outpatient/inpatient service use with variations between 23.2% (Denmark) and 79.1% (Poland) (Figure 1). This proportion was 47.9% for elevated specialist care and varied between 38.3% (the UK) and 66.4% (Poland). Around one-third (n=1,213; 35.9%) of the children had no elevated care use.

Risk of elevated outpatient/inpatient service use was associated with lower GA, BPD and other severe neonatal morbidities after adjustment for sociodemographic factors (Table 4). The risk was slightly lower for children moderately SGA (3rd–9th percentile). Children of mothers with intermediate educational level (compared to high) or unemployed parents at age five had higher risk of elevated outpatient/inpatient service use, whereas children of older mothers (≥35 years at delivery) had lower risk.

Elevated specialist service use was associated with low GA, BPD, congenital anomalies and other severe neonatal morbidities (Table 4). The risk was slightly higher for children moderately SGA (3–9th percentile). Boys had a higher risk of elevated specialist service use after adjustment for perinatal characteristics. Children of mothers with the lowest educational level at age five had slightly higher risk and non-European born mothers had lower risk of having elevated specialist service use.

Living in a country with higher routine follow-up coverage for children born VPT was associated with lower use of outpatient/inpatient services, but not with use of specialist services. Sensitivity analysis using the total number of outpatient/inpatient and specialist care contacts identified similar risk factors for elevated care (Table 5 [online]). Using inverse probability weights had limited impact on model estimates (see Table 6 [online] for models without weights).

**Discussion**

Children born VPT in the EPICE cohort were frequent users of health services during their fifth year of life, with wide variations between countries. Both elevated outpatient/inpatient and specialist service use were associated with lower GA, BPD and neonatal morbidities, with higher risk estimates for specialist service use. A higher risk of elevated specialist service use was also associated with moderate SGA status, congenital anomalies and male sex. In contrast, associations between a higher risk of service use and intermediate parental educational level, unemployment and a lower risk and higher maternal age at delivery were more pronounced for outpatient/inpatient services whereas the risk of having elevated specialist service use was lower for children of parents born outside Europe. Living in a country with high rates of routine follow-up for children born VPT was associated with lower risks of elevated outpatient/inpatient service use, but not specialist service use.

Strengths of this study include using data from a large, population-based cohort from regions in 11 European countries with comparable levels of development and universal health insurance. Comprehensive information was collected on health service use at five years of age using harmonized definitions that were developed by researchers in each country and refined based on parental responses at two years (8).Limitations include reliance on parent-report data and parents’ recall of the number of health-care visits over the past year, which may have led to an underestimation of service use; we were not able to validate service use against external sources. A systematic review on the validity of self-report questionnaires in adults showed that the accuracy of self-reported care visits varies depending on the services assessed and the patient population (46). Globally, results were inconsistent for GP visits across studies with both under and over-reporting, but other services, such as physiotherapy visits tended to be over-reported (46). Another study in adult patients found self-reported hospitalization and ER visits to be consistent with those extracted from medical records, but that GP visits were underreported (47). A study comparing administrative data to care-giver reported service use for children with asthma showed relatively high agreement over a two-year period, especially for hospitalizations, but found higher discrepancies for outpatient visits and higher discrepancies with increasing number of visits for all services (48). This literature suggests that any recall bias would likely lead to an underestimation of health service use, which would not call into question our finding of elevated service use. Further, other specialist services reported by some parents in free text responses were not included in the analysis to ensure consistency in ascertainment, leading to slight underestimation of service use. Future studies could use these responses to propose more comprehensive checklists. We did not have data on the reason for the health-care visits or measures of unmet health-care needs and could therefore not estimate the appropriateness of care or assess foregone health care. Finally, we used inverse probability weights to correct for study attrition, but this may not account for all sources of bias.

Our results show elevated health service use at five years of age in children born VPT, from multiple providers, including consultation with two or more specialists by 40.7% of the children. These results corroborate previous studies (1, 6) but contrast markedly with studies in the general population; for instance, 2017 and 2018 nationwide survey data from the U.S. show that approximately 13% of all children aged 0–17 years had elevated special health-care needs (49). The range of providers consulted illustrates the multiple concerns children may have as a consequence of VPT birth. The highest mean number of visits were reported for speech and physiotherapists, which can be expected for long-term interventions, but a smaller number of children with the most complex care needs have also contributed to this higher mean.

Elevated care use was consistently associated with perinatal risk factors, including lower GA, BPD and neonatal morbidities (IVH, cPVL, ROP and NEC). Elevated specialist service use was also associated with moderate SGA and congenital anomalies, and a stronger association with neonatal morbidities. This is expected, as children with perinatal risk factors are more likely to develop health and developmental difficulties requiring more, sometimes complex, care from multiple providers. Boys had a higher risk of elevated specialist care, which could be explained by their higher risks of neurodevelopmental delay (3).

Children with at least one unemployed parent and mothers with lower educational level at age five had higher risk of elevated outpatient/inpatient service use after adjusting for perinatal risk. Furthermore, although children with social disadvantage have an additional risk of poor developmental outcomes after adjusting for perinatal risk (5, 18, 19), use of specialist services in these children of mothers with the lowest educational level (RR=1.10) was not higher than for outpatient/inpatient care, (RR=1.14), and children with mothers born outside Europe were less likely to have specialist care. Previous research has found that high-risk children with additional social disadvantage are less likely to use medical services (24) and more likely to discontinue follow-up (21, 50). Poorer care access in socially disadvantaged populations may stem from inequitable referral by care providers (51), insufficient personal resources, lack of transportation, inflexible working conditions (21) or limited care provision in more deprived areas (27). We have previously shown that children with social risk factors have a higher risk of never using follow-up services (13), possibly leading to more restricted access to specialists and poorer care management resulting in higher reliance on ER and hospital health-care services (14, 30). Difficulties accessing specialized care, inappropriate or elevated use of ER services as well as social factors are modifiable risk factors for mortality and morbidity in children, that could be addressed with improved care coordination (52) and should be considered in follow-up programs.

Health service use varied between countries, as previously observed for specialist services at two years of age in this cohort (8), which can result from differences in the organization of primary pediatric care (53) and from differences in the accessibility of specialists and preventive services, including whether there is gate-keeping for specialist services and whether care provision is community or hospital based (54), as well as parents ability to pay out-of-pocket costs (55) if such costs exist. Recent studies have shown that care provision for children with complex care needs depends on their place of residence (30), and that there is a lack of consensus on how to meet complex care needs (29), which could explain variability in service use. Other differences in health-care systems as well as the society in general may play a role in the use of health care services, which were not able to account for in this study.

Although recent standards recommend multidisciplinary follow-up with coordinated and integrated care until five years of age, including sensory, neurodevelopmental, motor, speech, cognitive, socio-emotional and mental health assessments (9, 10), international consensus has not been established, with varying policies, for instance, in follow-up duration in Europe (11, 12). An overview of what follow-up is actually offered, and for how long, is currently lacking. Most children born very preterm are likely to be followed until two years of age, but longer follow-up varies across European countries (13).

The use of routine follow-up services at five years of age in the cohort was 27% overall, ranging from 11 to 58% across countries (13), which we hypothesized could have an impact on the use of health care services. The organization and subsequent use of follow-up services could impact the extent to which risk of neurodevelopmental sequelae is detected and treated. Our previous analyses using data from the cohort have shown high use of specialist services at two years’ corrected age (8), an age where the majority of children are still being followed up in programs for infants born VPT. We explored whether living in a country with more extensive follow-up for this population might affect health service use at five years of age, when follow-up is no longer systematic in all study regions and found that higher overall routine follow-up rates at five years were negatively associated with elevated outpatient/inpatient care use, but not elevated specialist care. However, there is no consensus on the content and duration of follow-up programs, and their implementation and the extent to which they correspond to policies remains unknown. In line with recent studies proposing that ER visits and hospitalizations may be avoided with improved care coordination (14, 15), our results suggest that well-established, longer and inclusive follow-up programs may help avoid undesirable health service use. This raises questions about equitable access to follow-up and health-care services in VPT populations and on the optimal organization of post-discharge care.

These differences across countries and between children based on socioeconomic or clinical risk raise the complex question of what constitutes optimal service use for children born VPT. Although it can be argued that specialist service use is more optimal than sick-visits, ER visits and hospitalizations, or no care at all, there is no definition on optimal care is in this population, and we cannot determine over or underuse without assessing service use in relation to care need. We were not able to assess the appropriateness of care used, but the lack of elevated specialist service use may be a sign of barriers to accessing needed care in subgroups where the rates of health and developmental problems are expected to be higher, such as in socially vulnerable families. Furthermore, interpreting the results is rendered difficult by the lack of comparable data on elevated health care service use in the general population. Recent European studies have shown that there is indeed variation in health care policies and care provision for children with complex care needs across Europe, however, there is little empirical data available that allow comparisons of health care services use across different populations. Future research should firstly collect representative and comparable data on elevated health care service use in the general population. Secondly, future studies are needed to investigate the reasons behind elevated service use and why it might vary across subgroups, for instance by using medical records to assess health and developmental problems and treatments received, as well as qualitative methods to explore family and provider perspectives on health service use in socially disadvantaged populations. Further, investigations in smaller, but well-defined sub-groups based on health status evaluated by standardized diagnostic tools, for instance children with cerebral palsy or severe cognitive, motor or sensorial impairments would generate more easily interpretable evidence about under and over-use of services.

**Conclusion**

This study provides previously unavailable data on types and amount of health-care services used at five years of age among children born VPT across regions in Europe. Our results reveal high service use, which varied across countries, and that elevated care use patterns may be different for outpatient/inpatient and specialist services, depending on perinatal and sociodemographic factors as well as the extent to which long-term follow-up is available. While many studies exist on developmental outcomes in children born VPT, our study emphasizes the importance of expanding research to investigate the services that should be provided to meet the long-term health needs of children born very preterm and to optimize health service use.

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**Figure legends**

**Figure 1.** Weighted proportionsa and 95% confidence intervals of elevated outpatient/inpatient and specialist service useb at five years of age in children born VPT, by country, ordered by overall routine follow-up rates

*Footnotes*

\*Rate of children still using routine follow-up services at five years below mean

aUsing inverse probability weights

bSee Table 2 for definitions