Short running head: Systematic review of economic consequences of preterm birth

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Abstract

Background: Despite extensive knowledge on the functional, neurodevelopmental, behavioural, and educational sequelae of preterm birth, relatively little is known about its economic consequences.

Objectives: To systematically review evidence around the economic consequences of preterm birth for the health services, for other sectors of the economy, for families and carers and, more broadly, for society.

Methods: Updating previous reviews, systematic searches of Medline, EconLit, Web of Science, the Cochrane Library, CINAHL, Embase and SCOPUS were performed using broad search terms, covering the literature from 1st January 2009 to 28th June 2017. Studies reporting economic consequences, published in the English language and conducted in a developed country were included. Economic consequences are presented in a descriptive manner according to study time horizon, cost category, and differential denominators (live births or survivors).

Results: Of 4,384 unique articles retrieved, 43 articles met the inclusion criteria. Of these, 27 reported resource use or cost estimates associated with the initial period of hospitalisation, whilst 26 reported resource use or costs incurred following the initial hospital discharge, 10 of which also reported resource use or costs associated with the initial period of hospitalisation. Only two studies reported resource use or costs incurred throughout the childhood years. Initial hospitalisation costs varied between $576,972 (range of $111,152 to $576,972) per infant born at 24 weeks' gestation to $930 (range of $930 to $7114) per infant born at term (US$, 2015 prices). The review also revealed a consistent inverse association between gestational age at birth and economic costs regardless of date of publication, country of publication, underpinning study design, follow-up period, age of assessment or costing

approach; and a paucity of evidence on non-healthcare costs. Several categories of economic costs, such as additional costs borne by families as a result of modifications to their everyday activities, are largely overlooked by this body of literature. Moreover, the number and coverage of economic assessments has not increased in comparison to previous review periods.

Conclusion: Evidence identified by this review can be used to inform clinical and budgetary service planning and act as data inputs into future economic evaluations of preventive or treatment interventions. Future research should focus particularly on valuing the economic consequences of preterm birth in adulthood.

Introduction

Preterm birth has been defined by the World Health Organisation (WHO) as any birth before 37 completed weeks' gestation, or fewer than 259 days since the first day of the mother's last menstrual period.[1] It has been further subdivided into subcategories based on gestational age at birth, including extremely preterm (<28 weeks' gestation), very preterm (28-<32 weeks' gestation), moderately preterm (32-<34 weeks' gestation) and late preterm (34-<37 weeks' gestation).[2] Almost 15 million babies worldwide were born preterm in 2010, representing 11.1% of all livebirths, with prevalence ranging from approximately 5% in several northern European countries to 18% in Malawi.[2] Moreover, time trends based on data from 65 countries with reliable time trend data between 1990 and 2010 and more than 10,000 live births per year reveal that the preterm birth rate has either remained stable (n=14) or increased (n=48) in 62 of those 65 countries.[2]

Mortality as a result of preterm birth complications ranks, together with pneumonia, as the leading cause of childhood deaths at the global level with over 3 million related annual neonatal deaths worldwide.[3] Although survival rates have improved in recent years, this masks considerable variation in the chances of survival across[4] and within[5] countries. Furthermore, surviving children are at higher risk of cerebral palsy, visual and auditory deficits, poor respiratory outcomes, impaired motor and cognitive ability and psychiatric disorders than children born at term.[6-9] Up to one third of children born extremely or very preterm and their parents face a life course with significant morbidity, dependency, and socioeconomic challenges.[10]

Despite an extensive body of knowledge on the functional, neurodevelopmental, behavioural, and educational sequelae of preterm birth,[11 12] relatively little is known about its economic consequences. This paper presents a systematic review of the recent scientific

literature on the economic consequences of preterm birth for the health services, for other sectors of the economy, for families and carers and, more broadly, for society. The paper represents an update of previous review articles of the topic published by one of the authors (SP) that had covered the periods 1980-1999 and January 2000 to June 2009, respectively.[13 14]

Methods

The systematic review followed the preferred reporting items for systematic reviews and meta-analyses (PRISMA) guidelines.[15] A comprehensive literature search strategy was developed and piloted, building upon our previous reviews of the topic.[13 14] The final search strategy encompassed granulated thesauri terms and free text search terms associated with preterm birth (e.g. preterm or premature* or low birthweight or gestational age) intersected with terms associated with economic consequences (e.g. "cost*" or "economic burden*" or "financial burden*" or "resource expenditure*" or "health care cost*" or "cost of illness"), and is presented in full in Appendix 1. The time horizon of the search strategy covered the period between 1st January 2009 and 28th June 2017. The electronic databases searched included Medline, EconLit, Web of Science (including Index to Scientific and Technical Proceedings, Science Citation Index, Social Science Citation Index), the Cochrane Library (including York Database of Abstracts of Reviews of Effectiveness, NHS Economic Evaluation Database), CINAHL, Embase and SCOPUS. Our search strategy was supplemented by manual reference searching of bibliographies, contacts with experts in the field, citation searching and author searching. All articles identified by the searches were entered into EndNote v7.7 and duplicates removed.

The main inclusion criteria framing the search strategy were primary studies reporting economic consequences associated with preterm birth (<37 weeks' gestation) or low

birthweight (<2500 g); published in the English language; and conducted in a developed country (defined, for the purposes of this review, as a member of the Organisation for Economic Co-operation and Development (OECD)). A two stage screening process was followed. Titles and abstracts were assessed at the first stage of the review by two independent reviewers (HHY and JK). If an article received approval from both reviewers, it proceeded to the next stage, with disagreements referred to a third reviewer (SP) for the final assessment. At the second stage, two reviewers (HHY and SP) independently reviewed full text articles to assess whether they met the review's inclusion criteria, with disagreements resolved via consensus. The term 'article' is used in its broadest sense and encompasses monographs, book chapters and conference abstracts. Studies of the economic consequences of low birth weight were included at the second stage of the review in order to capture potentially relevant information from contexts of clinical uncertainty in and incomplete recording of estimates of gestation. In addition, studies that reported the resource consequences associated with preterm birth or low birthweight in metrics amenable to future economic analyses were included at the second stage of the review for completeness. We excluded studies at the full article stage that: (i) were not relevant to economic aspects of preterm birth or low birthweight; (ii) did not report primary research evidence; (iii) were not published in the English language; (iv) treated preterm birth or low birthweight as a covariate rather than as a dependent variable or outcome; or (v) provided insufficient information to arrive at an economic assessment.

From each article that met the study selection criteria, we extracted the following information about the characteristics of the study using a bespoke proforma and entered it into an excel database: (i) bibliographic details, including year of publication; (ii) date of cohort/study; (iii) country/geographical jurisdiction; (iv) study design; (v) sample size(s); (vi) gestational age(s) at birth of study participants; (vii) birth weight(s) of study participants;

(viii) period over which study participants were followed up; (ix) cost categories; (x) resource use categories; (xi) currency in which costs were expressed; (xii) financial year for which costs were valued; and (xiii) cost data sources for the valuation process. The quality of contributing studies was assessed using a subset of 18 relevant items selected from the 24-item CHEERS reporting checklist for health economic evaluations.[16] Cost data extracted from studies was inflated, where necessary, to 2015 prices using the relevant country-specific Gross Domestic Product deflator index, and subsequently converted, where necessary, from their respective currencies into US dollars using purchasing power parities supplied by the OECD.[17] For studies that failed to report their currency price dates, it was assumed that the costs used in the valuation process applied to the financial year prior to the publication of the study.

Methodological variations between studies, including variations in underpinning health care practices across jurisdictions and variations in the relative prices of labour and capital inputs across jurisdictions, prevented a pooling of economic data akin to the meta-analyses performed on clinical effectiveness estimates. Rather, resource use and economic cost estimates are presented in a descriptive manner according to study time horizon, resource use and/or cost category, and differential denominators (live births or survivors).

Results

A total of 4,384 unique articles were identified by the combined literature searches (Figure 1). Of these, only 107 articles satisfied the first stage inclusion criteria, whilst a further 64 articles were excluded at the second stage of the review process. A total of 43 articles met the inclusion criteria at both stages and were included in the systematic review.[18-60] Of these 43 articles, 27 reported resource use or cost estimates associated with the initial period of

hospitalisation,[18-21 23-26 29 31 36-38 40 42-44 46-49 51 53 54 56 57 59] whilst 26 reported resource use or costs incurred following the initial hospital discharge,[21-23 25 27 28 30 32-36 38-41 45-47 50 52 54-56 58 60] 10 of which also reported resource use or costs associated with the initial period of hospitalisation.[21 23 25 36 38 40 46 47 54 56] Only two studies reported resource use or costs incurred throughout the childhood years.[33 46] The quality of studies that met the inclusion criteria of the review is summarised in Appendix 2 with quality assessment scores ranging from 2[24 44 48 51] and 17.[35 40 57] Notable methodological limitations included a failure to specify price dates for cost estimates,[18 21 23 34 43 44 47 48 51] failure to report discount rates for longer-terms costs,[22 30 33 34 50 54 55 58] and reliance on charges, which are likely to have included elements arising from corporate financial decisions.[19]

The methodological characteristics of studies reporting resource use and/or economic cost estimates associated with the initial hospitalisation, including their underpinning design, sample size, study population, study perspective and data sources are summarised in brief format in Table 1 and in full format in Appendix 3. Study estimates of hospital length of stay and economic costs are presented for differential denominators (live births or survivors) in brief format in Table 2 and in full format in Appendix 4. Initial hospitalisation costs varied between $576,972[29] (range of $111,152[57] to $576,972[29]) per infant born at 24 weeks' gestation to $930[43] (range of $930[43] to $7114[59]) per infant born at term, and from $169,132 per surviving infant born at <1,500g[51] to $1,200 per infant born at ≥2500g[21] (US$, 2015 prices). The duration of the initial hospitalisation varied between a median of 116[31] days (range of a mean of 84[53] to a median of 116[31] days) per surviving infant born at 24 weeks' gestation and a mean of 2.4[21] days (range of 2.4[21] to 5.2[42]) per infant born at term. A consistent inverse association was observed between gestational age at birth and economic costs regardless of date of publication, country of publication, underpinning study design or costing approach. The only exceptions were three studies that showed that, amongst infants

born at ≤26 weeks' gestation, costs increased as gestational age increased.[44 57 59] The denominators for these analyses were live births and therefore included infants that died as well as those that survived; the implication is that initial hospitalisation costs increased with gestational age in these cohorts as a consequence of the improved survival chances of the least immature. Only two studies estimated maternal costs to understand additional resources that may be associated with preterm birth.[29 47] Both studies revealed an inverse association between gestational age at birth and mean maternal costs. Moreover, only one study estimated non-healthcare costs associated with the initial period of hospitalisation.[25] In a prospective cohort study of 150 very low birth weight (VLBW) infants without prematurity-related morbidities and 145 full-term controls, Cavallo *et al*. used parental questionnaires to estimate that travel costs borne by parents and the value of lost productivity represented 7.2% and 29.6% of total societal costs, respectively, in the VLBW group, and 1.6% and 64.1% of total societal costs, respectively, in the control group.[25] Another notable feature of economic studies that focussed on the initial hospitalisation is the observed independent effect of multiplicity on increased hospitalisation costs.[43 57]

The methodological characteristics of studies reporting resource use and/or economic costs following initial hospitalisation are summarised in Appendix 5 with study estimates of economic outcomes presented in Appendix 6. Ten studies estimated outcomes during the first two years of life[21 23 25 28 36 41 45 47 56 60] with the remainder of studies estimating economic outcomes over longer follow-up periods or at later ages within cross-sectional studies. A consistent inverse association was observed between gestational age at birth and economic costs regardless of period of follow-up and age at assessment. This pattern held when children born moderate or late preterm were compared to children born at term,[22 23 27 30 36 41 47 55 60] and when children born at early term were compared to children born at late term.[30 41 55 60] Longitudinal studies with assessments of economic outcomes at repeated time points revealed

that health service resource use and costs declined as children aged, regardless of gestational age at birth.[27 30 38 50 54] Eight studies estimated economic costs accruing post initial discharge, associated with preterm birth, which are borne outside of the health sector.[25 30 35 36 39 46 47 52] Non-health service costs exceeded health service costs in the studies by Cavallo *et al*,[25] which was limited to an 18 month time horizon, and the study by Petrou *et al*,[52] which was limited to the 11th year of life, although no study valued economic costs borne by all sectors of the economy and by caregivers.

The two studies that reported economic costs throughout the childhood years used different methodological approaches. The earlier study, by Mangham *et al*,[46] applied decision-analytic modelling to estimate public sector costs associated with preterm birth. The model assumed a hypothetical cohort of children, equivalent in size to the number of live births in England and Wales in 2006, and was populated using data from thee cohort studies, namely EPICure, the Victorian Infant Collaborative Study, and the Oxford Record Linkage Study. The mean incremental public sector cost per preterm child surviving to 18 years, compared with a term survivor, was estimated at $39,329 (US$, 2015 prices). The corresponding estimates for a very and extremely preterm child were substantially higher at $106,174 and $162,816, respectively. More recently, Hummer *et al*[33] applied econometric methods to Austrian health insurance administrative panel data linked to birth registry data. They found that although absolute differences in health service utilisation between children with birthweights <2500g and ≥2500g diminished over time, those born at low birthweight still spent significantly more days in hospital and generated significantly higher medical drug and medical assistance expenses through early childhood. Furthermore, there was a shift during the years of compulsory schooling towards care for diseases of the nervous system and mental and behavioural disorders among children born at low birthweight with effects persisting until early adulthood.

Discussion

This paper systematically reviews the recent (2009-2017) scientific literature on the economic consequences of preterm birth for alternative stakeholders, updating two previous review articles of the topic.[13 14] The number of relevant studies identified by this review was smaller than the number of studies covered by the most recent review (43 vs 52), which covered a comparable time horizon, although the number of countries in which studies were conducted increased (13 vs 6).[14] In addition, a smaller proportion of studies estimated economic consequences following the initial hospital discharge (60.5% vs 67.3%) and economic consequences outside the health sector (18.6% vs 25.0%).[14] There does not appear to be evidence that the increased longevity of cohort studies of children born preterm or the establishment of collaborative research platforms around preterm birth (e.g. http://www.apic-preterm.org/) has increased the number or coverage of economic assessments.

When viewed in conjunction with evidence from previous systematic reviews,[13 14] the economic costs to the health services, other sectors of the economy and to families and caregivers associated with preterm birth remain considerable. Comparisons of cost estimates with those reported for other childhood conditions are constrained by a number of methodological factors, including differences in costing methodologies, ages at assessment, study perspectives and periods of follow-up, as well variations in underpinning health care practices and relative prices of resource inputs across study settings. Nevertheless, a holistic overview of the identified evidence suggests that ongoing economic costs associated with extremely preterm and very preterm birth exceed the annual cost burdens reported for several chronic childhood conditions, including asthma,[61] juvenile idiopathic arthritis,[62] depression,[63] separation anxiety[64] and attention-deficit hyperactivity disorder.[65]

Several categories of economic costs have, with a few exceptions,[36 52] been overlooked by this body of literature. These includes costs borne by local authorities and voluntary organisations, such as adaptations that have to be made to the individual's home as a result of their impaired state of health, and additional costs borne by families as a result of modifications to their everyday activities. In addition to the costs of travel, child care and accommodation, other potential costs faced by families and informal carers that have been largely overlooked include incremental expenditures on health goods, such as alternative therapies, and non-health goods, such as nutritional requirements, laundry, clothing, heating utilities and repairs to the home. Methods for ascertaining these costs, including questionnaires, diaries and data extraction from administrative databases, are available and widely applied in other areas of health care.[66] The value of ascertaining these costs should be balanced against the additional burden it may impose on study participants.

The full effects of impairment associated with preterm birth on economic outcomes clarifies only with time. Cohort studies with follow-up into adulthood and whole-country record linkage studies provide potential vehicles for ascertaining long-term economic outcomes such as long-term use of health and social care services, employment and occupational status, income, receipt of social welfare benefits and reproductive health, which might in turn have economic sequelae. As yet, no study has valued these long-term economic sequelae in adulthood. Recently established collaborative research platforms with a focus on outcomes in adulthood, such as the 'Adults Born Preterm International Collaboration' (http://www.apic-preterm.org/) and the 'Research on European Children and Adults born Preterm' programme (https://recap-preterm.eu/) should provide opportunities for economic research.

The strengths of this study are that it was conducted according to internationally agreed design and reporting guidelines for systematic reviews of economic evidence,[67] and was

broadly consistent in its methodology with previous reviews of the topic, thereby allowing inter-temporal comparisons to be made.[13 14] Limitations include exclusion of health utility-based measures of economic outcome that can be used to inform cost-effectiveness based decision-making.[68] It is likely that preterm birth is also associated with decrements in health utilities, i.e. preference-based health-related quality of life outcomes,[69] which have not been assessed in this review. Furthermore, the review did not extend to an assessment of the cost-effectiveness of preventive or treatment interventions for preterm birth.

The data identified by this review can be used to inform clinical and budgetary service planning and provide policy-relevant information for cross-country, longitudinal, and other cost comparisons.[70] The data can also act as inputs within cost-effectiveness models for preventive or treatment interventions. There are several circumstances, particularly in the context of decision-analytic modelling-based economic evaluations, where economic analysts lack the time and resources to collect primary data for economic parameters of interest. This review has generated a body of data that can act as inputs into future economic models, and allows the reader to assess the suitability of the evidence for their particular context.

In conclusion, this paper presents a systematic review of the recent scientific literature on the economic consequences of preterm birth for the health services, for other sectors of the economy, for families and carers and, more broadly, for society. Future research should focus particularly on valuing the economic consequences of preterm birth in adulthood.

What is already known on this topic

- There is extensive knowledge on the functional, neurodevelopmental, behavioural, and educational sequelae of preterm birth.
- Evidence on the economic consequences of preterm birth is more limited with previous reviews of the topic extending only to 2009.

What this study adds

- This systematic review of the 2009-2017 literature on the economic consequences of preterm birth reveals no evidence that the increased longevity of cohort studies of children born preterm or the establishment of collaborative research platforms around preterm birth has increased the number or coverage of economic assessments.
- The review reveals a consistent inverse association between gestational age at birth and economic costs regardless of date of publication, country of publication, underpinning study design, follow-up period, age of assessment or costing approach; and a continuing paucity of evidence on non-healthcare costs.
- Evidence identified by this review can be used to inform clinical and budgetary service planning and act as data inputs into future economic evaluations of preventive or treatment interventions.

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Contributorship statement

SP designed this study, reviewed the evidence from all contributing studies, wrote the article and acts as guarantor. HHY and JK screened all titles and abstracts. HHY also reviewed the evidence from all contributing studies.

Competing interests

None declared.

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Table 1: Studies reporting resource use and/or economic costs associated with the initial hospitalisation (full methodological characteristics presented in Appendix 3)

Study	Date of birth of study population	Location	Type of study	Sample size	Gestational age(s) (weeks)	Birth weight (g)
Allen *et al*[18]	2012-2015	USA	Case series	23 weeks: 12 24 weeks: 17 25 weeks: 22	23–25	<2,500
Almond *et al*[19]	1983-2002 1991-2006	USA 5 US states (Arizona, California, Maryland, New Jersey, and New York)	Retrospective analysis of national birth cohort State inpatient research database analysis	>200,000 newborns of three ounces around the 1,500 g threshold in the nationwide data; Approximately 30,000 births in the same interval for the five-state sample.	NA	3 ounces (85g) bandwith from 1,500g threshold
Aly *et al*[20]	2007-2008	USA	Retrospective analysis of national birth cohort	All: 81,913 33–34 weeks: 24,086 35–36 weeks: 57,827	33-34 35-36	NA
Barradas *et al*[21]	2009	USA	Retrospective analysis of national birth cohort	4,167,000 (9.1% were preterm or low birthweight)	<37	<2,500
Bird *et al*[23]	2001-2005	USA (Arkansas)	Retrospective analysis of Medicaid claims database	34-36 weeks: 5188 37-42 weeks: 15,303	34-36 37-42	NA
Bulut *et al*[24]	2006-2008	Turkey (Istanbul)	Retrospective chart analysis	34-36 weeks: 6742 ≥37: 30,055	34-36 ≥37	NA
Cavallo *et al*[25]	2006	Italy	Prospective cohort study	≤30 weeks and/or ≤1,500g: 150 37-42 weeks: 145	≤ 30 37-42	≤1,500

Comert *et al*[26]	2006-2007	Turkey (Istanbul)	Retrospective chart analysis	28-32 weeks: 96 33-37 weeks: 115	28-32 33-37	NA
Hall and Greenberg[29]	2012	USA (Hamilton County, Ohio)	Retrospective chart analysis	10,976 births of which 1444 were preterm	23-27 28-31 32-36	NA
Hinchliffe *et al*[31]	2006-2010	UK (East Midlands and Yorkshire)	Population-based audit of neonatal care	24 weeks: 296 25 weeks: 384 26 weeks: 551 27 weeks: 663 28 weeks: 829	24-28	NA
Khan *et al*[36]	2009-2010	UK (East Midlands)	Prospective cohort study	32-33 : 162 34-36: 984 32-36 weeks: 1146 ≥37 weeks: 1258	32-33 34-36 32-36 ≥37	NA
Khashu *et al*[37]	1999-2002	Canada (British Columbia)	Retrospective analysis of regional birth cohort	33–36 weeks: 6381 37–40 weeks: 88,867	33–36 37–40	NA
Korvenranta *et al*[38]	2000-2003	Finland	Retrospective national register study	22-23 weeks: 17 24-25 weeks: 135 26-27 weeks: 234 28-29 weeks: 391 30-31 weeks: 771 >31 weeks: 248	<32	<1501

Korvenranta *et al*[40]	2000-2003	Finland	Retrospective national register study	Preterm survivors: 23 weeks: 17 24-25 weeks: 135 26-27 weeks: 231 28-29 weeks: 385 30-31 weeks: 756 Preterm non-survivors: 278 Term: 200,609	<32 37–41	<1501
Leone *et al*[42]	2006-2007	Switzerland	Retrospective chart analysis	34-36 weeks: 530 39-40 weeks: 1686	34-36 39-40	NA
Lim *et al*[43]	2005-2006	Canada	Retrospective national register study	Singletons: <28 weeks: 762 28-31 weeks: 1099 32-33 weeks: 1428 34-36 weeks: 9716 ≥37 weeks: 189,750 Multiples: <28 weeks: 264 28-31 weeks: 418 32-33 weeks: 571 34-36 weeks: 2370 ≥37 weeks: 2742	<28 28-31 32-33 34-36 ≥37	NA
Lo *et al*[44]	2000-2008	USA	Retrospective chart analysis	Singleton deliveries: 240,179	24-42	NA
Mangham *et al*[46]	2006	England and Wales	Decision-analytic model populated with data from administrative population-based databases	≤ 27 weeks: 3185 28-31 weeks: 5575 32-33 weeks: 6410 34-36 weeks: 32,812 ≥37 weeks: 621,618	All	NA
McLaurin *et al*[47]	2004	USA	Retrospective analysis of insurance database	33-36 weeks: 1683 ≥37 weeks: 33,745	33–36 ≥37	NA
Meyers and Meyers[48]	2008	USA	Retrospective analysis of national birth cohort	Not specified	All	All

Mistry *et al*[49]	2001-2002	UK (Basildon, Colchester and Southend in Essex and Gillingham in Kent)	Prospective cohort study	Three level 2 neonatal units: 192 One level 3 neonatal unit: 199	NA	≤1,800
Nordermoen and Bratlid[51]	1997-2007	Norway	Retrospective analysis of two birth cohorts	Not specified	<28 weeks ≥28 weeks	<1500g
Seaton *et al*[53]	2011-2014	England	Retrospective analysis of electronic health records	2011: 5368 2012: 5343 2013: 5228 2014: 5099	24-31	NA
Stephens *et al*[54]	2001-2011	Australia (New South Wales)	Retrospective analysis of regional birth cohort	24-27 weeks: 3659 28-31 weeks: 7235 32-33 weeks: 9007 39-40 weeks: 539,631	24-27 28-31 32-33 39-40	NA
Thanh *et al*[56]	2004-2010	Canada (Alberta)	Retrospective analysis of regional birth cohort	<2,500g: 16,209 ≥2,500g: 189,586	NA	<2,500g ≥2,500g
van Baaren *et al*[57]	2006-2012	Netherlands	Retrospective analysis of one prospective cohort study and three randomised controlled trials	4,552 1090 singletons 3462 multiples	24–28 28–32 32–36 ≥37	NA
Xu *et al*[59]	2003	USA (Michigan)	Retrospective analysis of regional birth cohort	<37 weeks: 9780 ≥37 weeks: 101,484	≥20	NA

NA, not applicable.

Table 2: Summary of economic consequences associated with the initial hospitalisation (full results presented in Appendix 4)

Study	Date of birth of study population	Unit of analysis	Summary economic results
Allen *et al*[18]	2012-2015	Mean cost per survivor (US$, 2016 prices)[±]	23 weeks: 266,000±25,700 24 weeks: 239,900±34,450 25 weeks: 208,550±54,550
Almond *et al*[19]	1983-2002	Mean cost per live birth (US$, 2006 prices) (1US$2006=1.16US$2015)[#]	94,000 for the three ounces prior to the 1500g threshold, falling to 85,000 at three ounces after the threshold
Aly *et al*[20]	2007-2008	Mean length of stay per live birth (days)	33-34 weeks: 10.7% ≤3 days; 89.3% > 3 days 35-36 weeks: 56.0% ≤3 days; 44.0% > 3 days 33-36 weeks: 42.7% ≤3 days; 57.3% > 3 days
Barradas *et al*[21]	2009	Mean cost per live birth (US$, 2015 prices)[±]	Medicaid (Med); Commercial (Com); Self-pay (SP) Preterm, low birthweight: 16,200; 15,300; 8,000 <1500g: 57,000; 61,100; 24,000 1,500–2,499g: 12,300; 12,600; 8,200 >2,500g: 4600; 4,500; 3300 No stated birthweight: 11,000; 10,300; 2600 Term and normal birthweight: 1300; 1300; 1200
Bird *et al*[23]	2001-2005	Mean length of stay per live birth (days)	34-36 weeks: 2.61 37-42 weeks: 1.96
Bulut *et al*[24]	2006-2008	Mean length of stay per live birth (days)	34-36 weeks: 7 ≥37 weeks: 4
Cavallo *et al*[25]	2006	Mean cost per live birth (€, 2008 prices) (1€$2008=1.45US$2015)[#]	Hospital: ≤30 weeks and/or ≤1500g: 20,502; Term: 907 Societal: ≤30 weeks and/or ≤1500g: 32,460; Term: 2640
Comert *et al*[26]	2006-2007	Mean cost per live birth (US$, 2011 prices)[±] (1US$2011=1.06US$2015)[#]	28-32 weeks: 2575; 33-37 weeks: 1525
Hall and Greenberg[29]	2012	Mean cost per live birth (US$, 2012 prices) (1US$2012=1.04US$2015)[#]	23 weeks: 599,000; 24 weeks: 553,000; 25 weeks: 507,000; 26 weeks: 413,000; 27 weeks: 347,000; 28 weeks: 277,000; 29 weeks: 216,000; 30 weeks: 172,000; 31 weeks: 123,000; 32 weeks: 85,000; 33 weeks: 55,000; 34 weeks: 19,000; 35 weeks: 11,000; 36 weeks: 6000

Hinchliffe *et al*[31]	2006-2010	Median length of stay per survivor (days)	24 weeks: 116 25 weeks: 107 26 weeks: 93 27 weeks: 76 28 weeks: 65
Khan *et al*[36]	2009-2010	Mean cost per live birth (£, 2010-11 prices) (1£2011=1.51US$2015)[#]	32-33 weeks: Postnatal: 2207.13; Neonatal: 9226.81; Transfer: 61.64; Post-mortem: 22.97; Surgery: 51.34; Investigations: 58.72; Other: 194.66; Total: 11,628.60 34-36 weeks: Postnatal: 1948.73; Neonatal: 2497.97; Transfer: 12.24; Post-mortem: 2.52; Surgery: 46.13; Investigations: 20.5; Other: 81.40; Total: 4528.02 32-36 weeks: Postnatal: 1985.26; Neonatal: 3449.17; Transfer: 19.23; Post-mortem: 5.41; Surgery: 46.86; Investigations: 25.91; Other: 97.41; Total: 5532.62 ≥37 weeks: Postnatal: 1708.54; Neonatal: 146.45; Transfer: 0.02; Post-mortem: 1.86; Surgery: 0.81; Investigations: 6.76; Other: 9.45; Total: 1864.38
Khashu *et al*[37]	1999-2002	Mean length of stay per live birth (days)	33-36 weeks: 5.92 37-40 weeks: 2.38
Korvenranta *et al*[38]	2000-2003	Mean length of stay per survivor (days)	22-23 weeks: 136.7; 24-25 weeks: 122.1; 26-27 weeks: 93.7; 28-29 weeks: 67.4; 30-31 weeks: 47.6; >31 weeks: 42.3
Korvenranta *et al*[40]	2000-2003	Mean cost per survivor (€, 2008 prices) (1€$2008=1.23US$2015)[#]	23 (weeks): 147,398; 24-25 (weeks): 120,179; 26-27 (weeks): 88,188; 28-29 (weeks): 56,588; 30-31 (weeks): 35,147; All preterm: 54,104; Term: 1334
Leone *et al*[42]	2006-2007	Mean length of stay per live birth (days)	34 weeks: 13.6; 35 weeks: 12.1; 36 weeks: 7.0; Term: 5.2
Lim *et al*[43]	2005-2006	Mean cost per live birth (Can$, 2008 prices)[±] (1Can$2008=0.89US$2015)[#]	Singletons: <28 weeks: 84,235 (singletons); 90,123 (multiples) 28-31 weeks: 43,718 (singletons); 47,318 (multiples) 32-33 weeks: 19,463 (singletons; 21,388 (multiples) 34-36 weeks: 5047 (singletons); 6494 (multiples) Any preterm: 9233 (singletons); 12,479 (multiples) ≥37 weeks: 1050 (singletons); 1871 (multiples)
Lo *et al*[44]	2000-2008	Mean cost per live birth (US$, 2010 prices)[±] (1US$2010=1.08US$2015)[#]	24 weeks: 98,162; 25 weeks: 83,070; 26 weeks: 90,936; 27 weeks: 79,463; 28 weeks: 59,928; 29 weeks: 52,998; 30 weeks: 40,568; 31 weeks: 30,853; 32 weeks: 22,612; 33 weeks: 16,597; 34 weeks: 9740; 35 weeks: 5015; 36 weeks: 2413; 37 weeks: 1469; 38 weeks: 1070; 39 weeks: 994; 40 weeks: 1017; 41 weeks: 1058; 42 weeks: 1072
Mangham *et al*[46]	2006	Mean cost per survivor (£, 2006 prices) (1£2006=1.72US$2015)[#]	<28 weeks: Delivery: 1762; Neonatal: 84,453 <33 weeks: Delivery: 708; Neonatal: 57,726 <37 weeks: Delivery: 322; Neonatal: 21,066

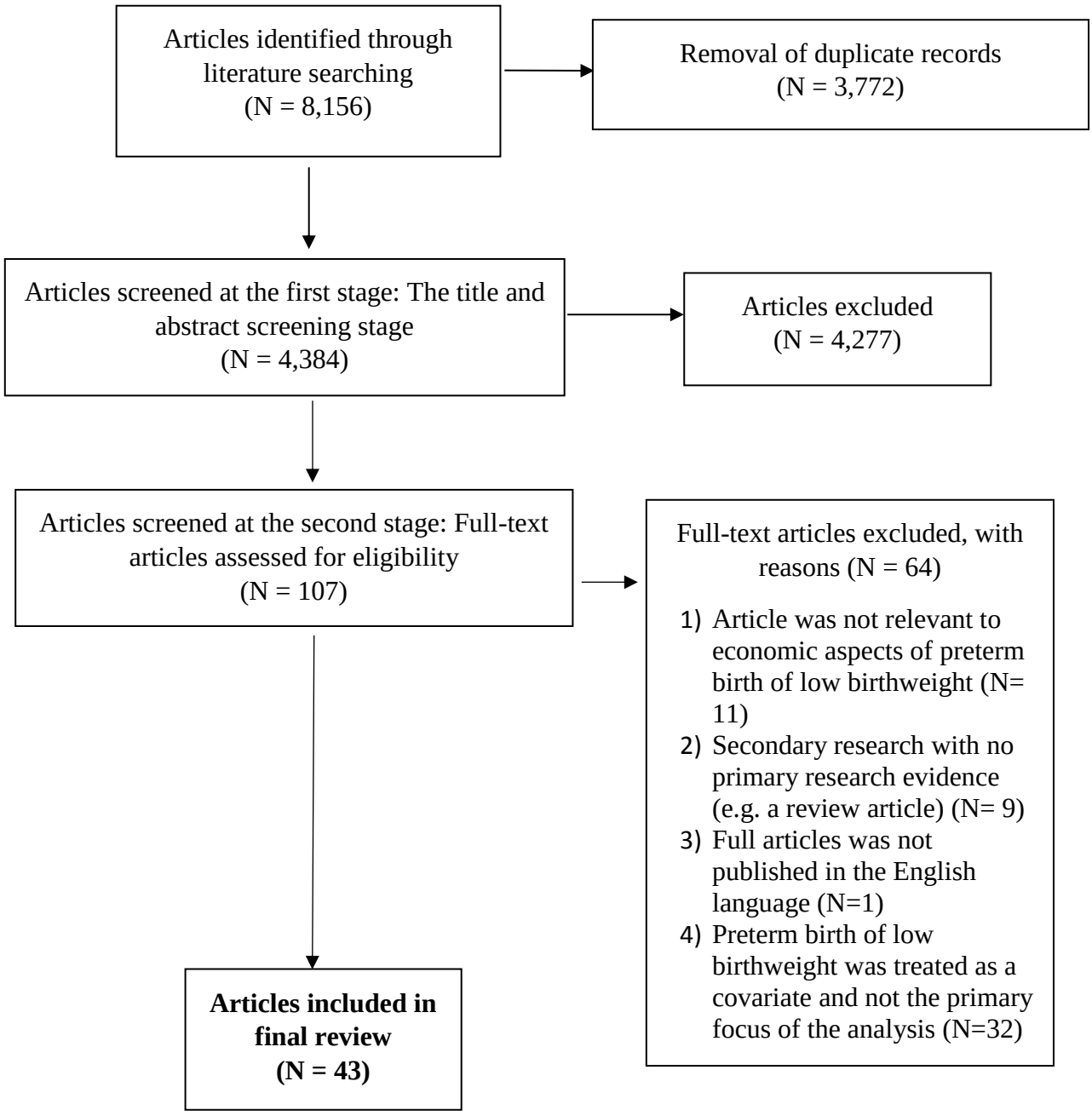
McLaurin *et al*[47]	2004	Mean cost per live birth (US$, 2008 prices)[±] (1US$2008=1.11US$2015)[#]	33-36 weeks: 26,054 ≥37 weeks: 2087
Meyers and Meyers[48]	2008	Mean cost per live birth (US$, 2010 prices)[±] (1US$2010=1.08US$2015)[#]	<2500g: 23,382 ≥2500g: 5951
Mistry *et al*[49]	2001-2002	Mean cost per live birth (£, 2005-6 prices) (1£2006=1.72US$2015)[#]	Excluding ambulance transfer costs: Level 2 units: 12,344; Level 3 unit: 17,861 Including ambulance transfer costs: Level 2 units: 12,881; Level 3 unit: 18,459
Nordermoen and Bratlid[51]	1997-2007	Mean cost per live birth/survivor (€, 2008 prices)[±] (1€$2008=1.30US$2015)[#]	<1500g: 1997: 67,700; 2007: 75.300/ <1500g: 1997: 130,100; 2007: 108,500 <28 weeks: 1997: 349,000; 2007: 429,500
Seaton *et al*[53]	2011-2014	Mean length of stay per live birth (days)	Intensive care: 24 weeks: 33; 25 weeks: 30; 26 weeks: 23; 27 weeks: 18; 28 weeks: 14; 29 weeks: 10; 30 weeks: 6; 31 weeks: 4 High dependency: 24 weeks: 29; 25 weeks: 33; 26 weeks: 29; 27 weeks: 25; 28 weeks: 19; 29 weeks: 13; 30 weeks: 9; 31 weeks: 6 Special care: 24 weeks: 22; 25 weeks: 27; 26 weeks: 30; 27 weeks: 31; 28 weeks: 33; 29 weeks: 33; 30 weeks: 31; 31 weeks: 28
Stephens *et al*[54]	2001-2011	Median cost per live birth (US$, 2014 prices) (1US$2014=1.01US$2015)[#]	24-27 weeks: 180,515 28-31 weeks: 62,450 32-33 weeks: 23,455 39-40 weeks: 2995
Thanh *et al*[56]	2004-2010	Mean cost per live birth (Can$, 2013 prices) (1Can$2013=0.83US$2015)[#]	<2500g: 18,126 ≥2500g: 1657
van Baaren *et al*[57]	2006-2012	Mean cost per live birth (€, 2011 prices) (1€$2011=1.26US$2015)[#]	<24 weeks: Singletons: NA; multiples: 41,700; 24 weeks: Singletons: 88,052; multiples: 79,572; 25 weeks: Singletons: 83,774; multiples:141,598; 26 weeks: Singletons: 71,821; multiples: 169,100; 27 weeks: Singletons: 70,984; multiples: 169,571; 28 weeks: Singletons: 60,783; multiples: 135,084; 29 weeks: Singletons: 45,444; multiples: 102,048; 30 weeks: Singletons: 34,510; multiples: 66,013; 31 weeks: Singletons: 26,296; multiples: 44,405; 32 weeks: Singletons: 21,157; multiples: 33,991; 33 weeks: Singletons: 15,912; multiples: 30,129; 34 weeks: Singletons: 11,222; multiples: 21,457; 35 weeks: Singletons: 6492; multiples: 14,306; 36 weeks: Singletons: 2924; multiples: 8618; ≥37 weeks: Singletons: 1434; multiples: 5201

Xu *et al*[59]	2003	Mean cost per live birth (US$, 2007 prices) (1US$2007=1.13US$2015)[#]	20 weeks: 11,397; 21 weeks: 11,703; 22 weeks: 25,367; 23 weeks: 48,908; 24 weeks: 99,477; 25 weeks: 82,296; 26 weeks: 103,980; 27 weeks: 72,212; 28 weeks: 56,933; 29 weeks: 45,598; 30 weeks: 34,642; 31 weeks: 29,679; 32 weeks: 24,623; 33 weeks: 21,887; 34 weeks: 18,617; 35 weeks: 15,864; 36 weeks: 12,305; ≥37 weeks: 6368

[±] As currency price dates not reported, it was assumed that the costs used in the valuation process applied to the financial year prior to study publication.

[#] Cost conversion rates reflecting relevant country-specific Gross Domestic Product deflator indices and purchasing power parities supplied by the OECD. These rates vary across individual countries using the same currency such as the €.[17]

Figure 1: PRISMA[#] Flow Diagram



[#] PRISMA denotes preferred reporting items for systematic reviews and meta-analyses.

Appendix 1: Literature search strategy

Search terms:

"gestational age" OR "low birthweight" OR "low birth weight" OR "low-birthweight" OR "low- birth-weight" OR "low-birth weight" OR ((pre-term OR preterm OR "pre term" OR prematur* OR early) **π** (birth OR delivery OR parturition OR labour OR labor OR "labour onset" OR "labor onset"))

AND

"cost*" OR "economic burden*" OR "financial burden*" OR "resource expenditure*" OR "health care cost*" OR "cost of illness" OR "costs and cost analys?s" OR "health expenditure*" OR "economic cost*" OR "economic analys?s" OR "economic consequence*" OR "economic implication*" OR "cost saving*" OR "medical expenditure*" OR "medical cost*" OR "healthcare expenditure*" OR "health care expenditure*" OR "healthcare cost*" OR "health care cost*" OR "cost allocation" OR "direct service cost*" OR "drug cost*" OR "employer health cost*" OR "hospital cost*" OR "capital expenditure*" OR "direct cost*" OR "indirect cost*" OR "intangible cost*".

Search engines: Medline, EconLit, Web of Science (including Index to Scientific and Technical Proceedings, Science Citation Index, Social Science Citation Index), the Cochrane Library (including York Database of Abstracts of Reviews of Effectiveness, NHS Economic Evaluation Database), CINAHL, Embase and SCOPUS.

Note: Medline, Embase and CINAHL required the incorporation of MeSH (Medical Subject Headings) terms during the literature search process. The MeSH terms of these three databases are summarised as follows:
Medline: "health expenditures", "costs and cost analysis" OR "cost of illness" OR "health care costs", "economics, hospital" AND "economics, medical", "cost saving", "cost allocation"; Embase: "cost" OR "drug cost" OR "hospital running cost" OR "hospital cost" OR "cost of illness" OR "hospitalization cost", "health care cost";
CINAHL: "health care costs" OR "costs and cost analysis" OR "health facility costs" OR "cost savings" OR "economic aspects of illness".

Also,

$$\boldsymbol{\pi} = \begin{cases} \text{adjn}, & \text{for Medline} \\ \text{Nn}, & \text{for Embase, EconLit and CINAHL} \\ \text{w/n}, & \text{for Scopus} \\ \text{NEAR/n}, & \text{for Web of Science, Cochrane Library} \end{cases}$$

where $n =$ the specified gap in number of words. Under the consideration of the above search terms, n was set to 3.

Time horizon: 1st January 2009 to 28th June 2017.

Appendix 2: Reporting quality assessment results according to a subset of the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) reporting items

Studies only reporting resource utilisation	
Study	Reporting quality score (out of maximum 16)
Aly *et al*[20]	14
Corchia *et al*[27]	14
Hinchliffe *et al*[31]	13
Hong *et al*[32]	13
Khashu *et al*[37]	13
Korvenranta *et al*[38]	14
Kuzniewicz *et al*[41]	12
Leone *et al*[42]	12
Luu *et al*[45]	13
Seaton *et al*[53]	14
Young *et al*[60]	13
Mean score	**13.18**
Studies reporting economic costs	
Study	Reporting quality score (out of maximum 18)
Allen *et al*[18]	12
Almond *et al*[19]	15
Barradas *et al*[21]	12
Berard *et al*[22]	16
Bird *et al*[23]	13
Bulut *et al*[24]	2
Cavallo *et al*[25]	16
Comert *et al*[26]	13
Gareau *et al*[28]	14
Hall and Greenberg[29]	16
Helle *et al*[30]	16
Hummer *et al*[33]	13
Jacob *et al*[34]	13
Johnston *et al*[35]	17
Khan *et al*[36]	16
Korvenranta *et al*[39]	16
Korvenranta *et al*[40]	17
Lim *et al*[43]	12
Lo *et al*[44]	2
Mangham *et al*[46]	16
McLaurin *et al*[47]	14
Meyers and Meyers[48]	2
Mistry *et al*[49]	14
Mourani *et al*[50]	15
Nordermoen and Bratlid[51]	2
Petrou *et al*[52]	14

Stephens *et al*[54]	16
Stephens *et al*[55]	15
Thanh *et al*[56]	15
van Baaren *et al*[57]	17
Westrupp *et al*[58]	16
Xu *et al*[59]	14
Mean score	**13.16**

Appendix 3: Full methodological characteristics of studies reporting resource use and/or economic costs associated with the initial hospitalisation

Study	Date of birth of study population	Location	Type of study	Sample size	Gestational age(s) (weeks)	Birth weight (g)	Type of resource inputs/costs	Data sources
Allen *et al*[18]	2012-2015	USA	Case series	23 weeks: 12 24 weeks: 17 25 weeks: 22	23–25	<2,500	Hospital	A single tertiary care, university-based NICU over 42 months
Almond *et al*[19]	1983-2002 1991-2006	USA, 5 US states (Arizona, California, Maryland, New Jersey, and New York)	Retrospective analysis of national birth cohort State inpatient research database analysis	>200,000 newborns of three ounces around the 1,500 g threshold in the nationwide data; Approximately 30,000 births in the same interval for the five-state sample.	NA	3 ounces (85g) bandwith from 1,500g threshold	Hospital (charges)	(i) Census of U.S. births with linked death and hospital discharge data; (ii) State-wide births linked to death and hospital discharge data.
Aly *et al*[20]	2007-2008	USA	Retrospective analysis of national birth cohort	All: 81,913 33–34 weeks: 24,086, 35–36 weeks: 57,827	33-34 35-36	NA	Hospital	US Nationwide Inpatient Sample Database
Barradas *et al*[21]	2009	USA	Retrospective analysis of national birth cohort	4,167,000 (9.1% were preterm or low birthweight)	<37	<2,500	Hospital	US Nationwide Inpatient Sample Database
Bird *et al*[23]	2001-2005	USA (Arkansas)	Retrospective analysis of Medicaid	34-36 weeks: 5188 37-42 weeks: 15,303	34-36 37-42	NA	Hospital	Arkansas Medicaid claims data linked to state birth certificate data

			claims database					
Bulut *et al*[24]	2006-2008	Turkey (Istanbul)	Retrospective chart analysis	34-36 weeks: 6742 ≥37: 30,055	34-36 ≥37	NA	Hospital	Zeynep Kamil Maternity and Childrens' Disease Training and Research State Hospital
Cavallo *et al*[25]	2006	Italy	Prospective cohort study	≤30 weeks and/or ≤1,500g: 150 37-42 weeks: 145	≤ 30 37-42	≤1,500	Hospital and broader societal	Prospective data collection in 25 neonatal intensive care units and from parent hospitals for controls. Parental questionnaires.
Comert *et al*[26]	2006-2007	Turkey (Istanbul)	Retrospective chart analysis	28-32 weeks: 96 33-37 weeks: 115	28-32 33-37	NA	Hospital	Dr. L. Kırdar Kartal Research and Training Hospital NICU
Hall and Greenberg[29]	2012	USA (Hamilton County, Ohio)	Retrospective chart analysis	10,976 births of which 1444 were preterm	23-27 28-31 32-36	NA	Hospital	Ohio Department of Health
Hinchliffe *et al*[31]	2006-2010	UK (East Midlands and Yorkshire)	Population-based audit of neonatal care	24 weeks: 296 25 weeks: 384 26 weeks: 551 27 weeks: 663 28 weeks: 829	24-28	NA	Hospital (neonatal care)	The Neonatal Survey: A population-based audit of inpatient neonatal care
Khan *et al*[36]	2009-2010	UK (East Midlands)	Prospective cohort study	32-33 : 162 34-36: 984 32-36 weeks: 1146 ≥37 weeks: 1258	32-33 34-36 32-36 ≥37	NA	Postnatal, neonatal, transfers, post-mortems, surgery, investigations.	Bespoke questionnaires designed for The Late and Moderate Preterm Birth Study (LAMBS)
Khashu *et al*[37]	1999-2002	Canada (British Columbia)	Retrospective analysis of regional birth cohort	33–36 weeks: 6381 37–40 weeks: 88,867	33–36 37–40	NA	Hospital	British Columbia (BC) Perinatal Database Registry and Reporting Tool of the BC Reproductive Care Program

Korvenranta *et al*[38]	2000-2003	Finland	Retrospective national register study	22-23 weeks: 17 24-25 weeks: 135 26-27 weeks: 234 28-29 weeks: 391 30-31 weeks: 771 >31 weeks: 248	<32	<1501	Hospital	Finnish Medical Birth Register and Finnish Hospital Discharge Register
Korvenranta *et al*[40]	2000-2003	Finland	Retrospective national register study	Preterm survivors: 23 weeks: 17 24-25 weeks: 135 26-27 weeks: 231 28-29 weeks: 385 30-31 weeks: 756 Preterm non-survivors: 278 Term: 200,609	<32 37–41	<1501	Hospital	Finnish Medical Birth Register and Finnish Hospital Discharge Register
Leone *et al*[42]	2006-2007	Switzerland	Retrospective chart analysis	34-36 weeks: 530 39-40 weeks: 1686	34-36 39-40	NA	Hospital	University Hospital of Zurich and its four surrounding hospitals: Zollikerberg, Triemli, Limmattal and Uster.
Lim *et al*[43]	2005-2006	Canada	Retrospective national register study	Singletons: <28 weeks: 762 28-31 weeks: 1099 32-33 weeks: 1428 34-36 weeks: 9716 ≥37 weeks: 189,750 Multiples: <28 weeks: 264 28-31 weeks: 418 32-33 weeks: 571 34-36 weeks: 2370 ≥37 weeks: 2742	<28 28-31 32-33 34-36 ≥37	NA	Hospital	Discharge Abstract Database and Canadian Management Information Systems Database, Canadian Institute for Health Information
Lo *et al*[44]	2000-2008	USA	Retrospective chart analysis	Singleton deliveries: 240,179	24-42	NA	Hospital	18 hospitals within a single health care system

Mangham *et al*[46]	2006	England and Wales	Decision-analytic model populated with data from administrative population-based databases	≤ 27 weeks: 3185 28-31 weeks: 5575 32-33 weeks: 6410 34-36 weeks: 32,812 ≥37 weeks: 621,618	All	NA	Delivery, neonatal	Secondary data from external cohorts and cross-sectional studies
McLaurin *et al*[47]	2004	USA	Retrospective analysis of insurance database	33-36 weeks: 1683 ≥37 weeks: 33,745	33–36 ≥37	NA	Hospital	MedStat MarketScan Commercial Claims and Encounters database
Meyers and Meyers[48]	2008	USA	Retrospective analysis of national birth cohort	Not specified	All	All	Hospital	2008 Healthcare Cost and Utilization Project Nationwide Inpatient Sample
Mistry *et al*[49]	2001-2002	UK (Basildon, Colchester and Southend in Essex and Gillingham in Kent)	Prospective cohort study	Three level 2 neonatal units: 192 One level 3 neonatal unit: 199	NA	≤1,800	Neonatal care	One regional level 3 unit and three level 2 units providing short-term intensive care
Nordermoen and Bratlid[51]	1997-2007	Norway	Retrospective analysis of two birth cohorts	Not specified	<28 weeks ≥28 weeks	<1500g	Hospital	Patient records
Seaton *et al*[53]	2011-2014	England	Retrospective analysis of electronic health records	2011: 5368 2012: 5343 2013: 5228 2014: 5099	24-31	NA	Neonatal care	UK National Neonatal Research Database

Stephens *et al*[54]	2001-2011	Australia (New South Wales)	Retrospective analysis of regional birth cohort	24-27 weeks: 3659 28-31 weeks: 7235 32-33 weeks: 9007 39-40 weeks: 539,631	24-27 28-31 32-33 39-40	NA	Hospital	Linked New South Wales Perinatal Data Collection, Admitted Patient Data Collection, and Registry of Births, Deaths and Marriages
Thanh *et al*[56]	2004-2010	Canada (Alberta)	Retrospective analysis of regional birth cohort	<2,500g: 16,209 ≥2,500g: 189,586	NA	<2,500g ≥2,500g	Hospital, NICU	Alberta Perinatal Health Program administrative databases
van Baaren *et al*[57]	2006-2012	Netherlands	Retrospective analysis of one prospective cohort study and three randomised controlled trials	4,552 1090 singletons 3462 multiples	24–28 28–32 32–36 ≥37	NA	Hospital	One prospective cohort study (APOSTEL I) and three randomised controlled trials (APOSTEL II, AMPHIA and PROTWIN)
Xu *et al*[59]	2003	USA (Michigan)	Retrospective analysis of regional birth cohort	<37 weeks: 9780 ≥37 weeks: 101,484	≥20	NA	Hospital	Linked live birth certificate files, hospital discharge data and mortality data from the Michigan Department of Community Health and the National Center for Health Statistics

NA, not applicable.

Appendix 4: Resource use and/or economic costs associated with the initial hospitalisation

Study	Currency	Price date	Cost per live birth (mean)	Cost per survivor (mean)	Maternal cost per live birth (mean)	Length of stay (days) per live birth (mean)	Length of stay (days) per survivor (mean)
Allen *et al*[18]	US $	Not specified	NA	23 weeks: 266,000±25,700 24 weeks: 239,900±34,450 25 weeks: 208,550±54,550	NA	NA	114±11 111±13 91±23
Almond *et al*[19]	US $	2006	94,000 for the three ounces prior to the 1500g threshold, falling to 85,000 at three ounces after the threshold	NA	NA	27.3 immediately prior to threshold; 25.7 immediately after the threshold.	NA
Aly *et al*[20]	NA	NA	NA	NA	NA	33-34 weeks: 10.7% ≤3 days 89.3% > 3 days 35-36 weeks: 56.0% ≤3 days 44.0% > 3 days 33-36 weeks: 42.7% ≤3 days 57.3% > 3 days	NA
Barradas *et al*[21]	US $	Not specified	Medicaid (Med); Commercial (Com); Self-pay (SP) Preterm, low birthweight: 16,200; 15,300; 8,000 <1500g: 57,000; 61,100; 24,000 1,500–2,499g: 12,300; 12,600; 8,200 >2,500g: 4600; 4,500; 3300 No stated birthweight: 11,000; 10,300; 2600 Term and normal birthweight: 1300; 1300; 1200	NA	NA	Med; Com; SP 12.8; 11.9; 7.2 38.3; 39.4; 15.9 10.9; 11.1; 7.9 4.6; 4.5; 3.9 11.0; 10.5; 1.7 2.4; 2.4; 2.3	NA

Bird *et al*[23]	NA	NA	NA	NA	NA	34-36: 2.61 37-42: 1.96	NA
Bulut *et al*[24]	NA	NA	NA	NA	NA	34-36: 7 ≥37: 4	
Cavallo *et al*[25]	€	2008	Hospital: ≤30 weeks and/or ≤1500g: 20,502; Term: 907 Societal: ≤30 weeks and/or ≤1500g: 32,460; Term: 2640	NA	NA	≤30 weeks and/or ≤1500g: 59.7 37-42 weeks: 3.0	NA
Comert *et al*[26]	US $	NA	28-32 weeks: 2575 33-37 weeks: 1525	NA	NA	28-32 weeks: 15.58 33-37 weeks: 12.04	NA
Hall and Greenberg[29]	US $	2012	23 weeks: 599,000 24 weeks: 553,000 25 weeks: 507,000 26 weeks: 413,000 27 weeks: 347,000 28 weeks: 277,000 29 weeks: 216,000 30 weeks: 172,000 31 weeks: 123,000 32 weeks: 85,000 33 weeks: 55,000 34 weeks: 19,000 35 weeks: 11,000 36 weeks: 6000	NA	9000 9000 9000 8000 8000 8000 8000 8000 7000 6000 5000 4000 4000 4000	NA	NA
Hinchliffe *et al*[31]	NA	NA	NA	NA	NA		24 weeks: 116* 25 weeks: 107* 26 weeks: 93* 27 weeks: 76*

							28 weeks: 65*
Khan *et al*[36]	£	2010-11	32-33 weeks: Postnatal: 2207.13; Neonatal: 9226.81; Transfer: 61.64; Post-mortem: 22.97; Surgery: 51.34; Investigations: 58.72; Other: 194.66; Total: 11,628.60 34-36 weeks: Postnatal: 1948.73; Neonatal: 2497.97; Transfer: 12.24; Post-mortem: 2.52; Surgery: 46.13; Investigations: 20.5; Other: 81.40; Total: 4528.02 32-36 weeks: Postnatal: 1985.26; Neonatal: 3449.17; Transfer: 19.23; Post-mortem: 5.41; Surgery: 46.86; Investigations: 25.91; Other: 97.41; Total: 5532.62 ≥37 weeks: Postnatal: 1708.54; Neonatal: 146.45; Transfer: 0.02; Post-mortem: 1.86; Surgery: 0.81; Investigations: 6.76; Other: 9.45; Total: 1864.38	NA	NA	NA	NA
Khashu *et al*[37]	NA	NA	NA	NA	NA	33-36 weeks: 5.92 37-40 weeks: 2.38	NA
Korvenranta *et al*[38]	NA	NA	NA	NA	NA	NA	22-23 (weeks): 136.7; 24-25: 122.1; 26-27: 93.7; 28-29: 67.4; 30-31: 47.6; >31: 42.3
Korvenranta *et al*[40]	€	2008	NA	23 (weeks): 147,398 24-25: 120,179 26-27: 88,188 28-29: 56,588	NA	NA	23 (weeks): 136.7 24-25: 122.1

				30-31: 35,147 All preterm: 54,104 Term: 1334			26-27: 94.4 28-29: 67.5 30-31: 48.0 All preterm: 64.4 Term: 3.5
Leone *et al*[42]	NA	NA	NA	NA	NA	34 weeks: 13.6 35 weeks: 12.1 36 weeks: 7.0 Term: 5.2	NA
Lim *et al*[43]	Can $	Not specified	Singletons: <28 weeks: 84,235 (singletons); 90,123 (multiples) 28-31 weeks: 43,718 (singletons); 47,318 (multiples) 32-33 weeks: 19,463 (singletons; 21,388 (multiples) 34-36 weeks: 5047 (singletons); 6494 (multiples) Any preterm: 9233 (singletons); 12,479 (multiples) ≥37 weeks: 1050 (singletons); 1871 (multiples)	NA	NA	Singletons: <28 weeks: 83.1 28-31 weeks: 42.6 32-33 weeks: 21.2 34-36 weeks: 5.8 Any preterm: 10.0 ≥37 weeks: 2.1 Multiples: <28 weeks: 91.0 28-31 weeks: 46.0 32-33 weeks: 23.5 34-36 weeks: 8.0 Any preterm: 13.8 ≥37 weeks: 3.5	NA
Lo *et al*[44]	US $	Not specified	24 weeks: 98,162 25 weeks: 83,070 26 weeks: 90,936 27 weeks: 79,463 28 weeks: 59,928 29 weeks: 52,998 30 weeks: 40,568 31 weeks: 30,853	NA	NA	NA	NA

			32 weeks: 22,612 33 weeks: 16,597 34 weeks: 9740 35 weeks: 5015 36 weeks: 2413 37 weeks: 1469 38 weeks: 1070 39 weeks: 994 40 weeks: 1017 41 weeks: 1058 42 weeks: 1072				
Mangham *et al*[46]	£	2006	NA	<28 weeks: Delivery: 1762 Neonatal: 84,453 <33 weeks: Delivery: 708 Neonatal: 57,726 <37 weeks: Delivery: 322 Neonatal: 21,066	NA	NA	NA
McLaurin *et al*[47]	US $	Not specified	33-36 weeks: 26,054 ≥37 weeks: 2087	NA	33-36 weeks: 6672 ≥37 weeks: 1943	33-36 weeks: 8.8 ≥37 weeks: 2.2	NA
Meyers and Meyers[48]	US $	Not specified	<2500g: 23,382 ≥2500g: 5951	NA	NA	<2,500g or <37 weeks: 11.9 Infants with an uncomplicated newborn stay: 2.3 All other infants: 4.2	NA

Mistry *et al*[49]	£	2005-6	Excluding ambulance transfer costs: Level 2 units: 12,344 Level 3 unit: 17,861 Including ambulance transfer costs: Level 2 units: 12,881 Level 3 unit: 18,459	NA	NA	Level 2 units: 29.8 Level 3 unit: 29.4	NA
Nordermoen and Bratlid[51]	€	Not specified	<1500g: 1997: 67,700 2007: 75.300	<1500g: 1997: 130,100 2007: 108,500 <28 weeks: 1997: 349,000 2007: 429,500	NA	NA	NA
Seaton *et al*[53]	NA	NA	NA	NA	NA	Intensive care: 24 weeks: 33; 25: 30; 26: 23; 27: 18; 28: 14; 29: 10; 30: 6; 31: 4 High dependency: 24 weeks: 29; 25: 33; 26: 29; 27: 25; 28: 19; 29: 13; 30: 9; 31: 6 Special care: 24 weeks: 22; 25: 27; 26: 30; 27: 31; 28: 33; 29: 33; 30: 31; 31: 28	NA
Stephens *et al*[54]	US $	2014	24-27: 180,515* 28-31: 62,450* 32-33: 23,455* 39-40: 2995*	NA	NA	24-27: 87* 28-31: 47* 32-33: 25* 39-40: 3*	NA
Thanh *et al*[56]	Can $	2013	<2500g: 18,126 ≥2500g: 1657	NA	NA	Hospital days: <2500g: 12.3 ≥2500g: 1.8 NICU days: <2500g: 11.2 ≥2500g: 0.2	NA

van Baaren *et al*[57]	€	2011	<24 weeks: Singletons: NA; multiples: 41,700 24 weeks: Singletons: 88,052; multiples: 79,572 25 weeks: Singletons: 83,774; multiples:141,598 26 weeks: Singletons: 71,821; multiples: 169,100 27 weeks: Singletons: 70,984; multiples: 169,571 28 weeks: Singletons: 60,783; multiples: 135,084 29 weeks: Singletons: 45,444; multiples: 102,048 30 weeks: Singletons: 34,510; multiples: 66,013 31 weeks: Singletons: 26,296; multiples: 44,405 32 weeks: Singletons: 21,157; multiples: 33,991 33 weeks: Singletons: 15,912; multiples: 30,129 34 weeks: Singletons: 11,222; multiples: 21,457 35 weeks: Singletons: 6492; multiples: 14,306 36 weeks: Singletons: 2924; multiples: 8618 ≥37 weeks: Singletons: 1434; multiples: 5201	NA	NA	NA	NA
Xu *et al*[59]	US $	2007	20 weeks: 11,397 21 weeks: 11,703 22 weeks: 25,367 23 weeks: 48,908 24 weeks: 99,477 25 weeks: 82,296 26 weeks: 103,980	NA	NA	NA	NA

			27 weeks: 72,212 28 weeks: 56,933 29 weeks: 45,598 30 weeks: 34,642 31 weeks: 29,679 32 weeks: 24,623 33 weeks: 21,887 34 weeks: 18,617 35 weeks: 15,864 36 weeks: 12,305 ≥37 weeks: 6368				

* Median value. NA, not applicable.

Appendix 5: Methods of studies reporting resource use and/or economic costs following initial hospital discharge

Study	Date of study popu-lation	Location	Type of study	Sample size	Gestational age(s) (weeks)	Birth weight (g)	Type of resource inputs/costs	Data sources	Time frame
Barradas *et al*[21]	2009	USA	Retrospective analysis of national birth cohort	4,167,000 (9.1% were preterm or low birthweight)	<37	<2500	Hospital	US Nationwide Inpatient Sample Database	First 28 days of life
Berard *et al*[22]	1997-2000	Canada (Quebec)	Retrospective cohort study	Term live births: 33,879 Late preterm live births: 2176	33-36 ≥37	NA	Hospitalisation, physician visits, emergency department visits and prescriptions following initial hospitalisation	Linkage of three Quebec administrative databases: Régie de l'Assurance Maladie du Québec, MED-ECHO, Institut de la Statistique du Québec	First 3 years of life; first 2 years of life; and third year of life
Bird *et al*[23]	2001-2005	USA (Arkansas)	Retrospective analysis of Medicaid claims database	34-36 weeks: 5188 37-42 weeks: 15,303	34 -36 37– 42	NA	Hospital inpatient and outpatient care	Arkansas Medicaid claims data linked to state birth certificate data	First year of life
Cavallo *et al*[25]	2006	Italy	Prospective cohort study	Gestational age ≤30 and/or birthweight ≤1500: 150 37-42 weeks: 145	≤ 30 37-42	≤ 1500	Hospital readmissions, outpatient visits, general practitioner consultations, broader societal	Parent questionnaires competed at 6 and 18 months	From initial hospital discharge to 18 months

Corchia *et al*[27]	2007-2008	Italy (Lazio)	Retrospective cohort study	90,545 of whom 628 were VPT, 6143 MLPT, 29,219 ET and 54,555 FT	VPT: 24-31 MLPT: 32-36 ET: 37-38 FT: 39-41	NA	Emergency department visits and re-hospitalisations	Linkage between the regional Birth Certification Register, the regional Hospital Discharge Register and the regional Emergency Department Register	Three years following initial hospital discharge
Gareau *et al*[28]	2009-2013	USA	Retrospective cohort study	1262 women involved in 'Centering Pregnancy' group prenatal care and 5066 receiving individual prenatal care	Low risk women	Low risk women	Professional and inpatient Medicaid claims	Medicaid data at a pilot programme in South Carolina	First year postpartum
Helle *et al*[30]	2006-2008	Finland (Municipalities of Helsinki, Espoo, Vantaa)	Prospective cohort study	<32 weeks: 198 32-33 weeks: 205 34-36 weeks: 1164 37-38 weeks: 4586 39-41 weeks: 21,812 >41 weeks: 2005	<32 32-33 34-36 37-38 39-41 >41	NA	Specialized care, primary care, private health care, medications	Linkage between national medical birth register and outpatient, inpatient, and primary care registers	First 3 years of life
Hong *et al*[32]	2000-2004	Australia (New South Wales)	Retrospective cohort study	<32 weeks: 2939 survivors	<32	NA	Respiratory-related hospitalisation	Linkage between New South Wales (NSW) Admitted Patient Data Collection and the NSW Registry of Births, Deaths and Marriages	First 3 years post initial hospital discharge

Hummer *et al*[33]	2005-2009	Austria	Econometric analysis of administrative data	<1500g: 119 (year 0-1) 462 (years 2-5) 1136 (years 6-14) 425 (years 15-21) <2500g: 854 (year 0-1) 2957 (years 2-5) 7926 (years 6-14) 4856 (years 15-21) ≥2500g: 18,579 (year 0-1) 70,328 (years 2-5) 227,548 (years 6-14) 134,739 (years 15-21)	NA	<1500 <2500 ≥2500	Outpatient expenditures, medical attendance, drugs	Administrative data from the Upper Austrian Sickness Fund and the Austrian birth register	Stratified ages: year 0-1 years 2-5 years 6-14 years 15-21
Jacob *et al*[34]	2011-2012	Germany	Retrospective cohort study	<28 weeks: 23 29-36 weeks: 526 ≥37 weeks: 5,398	<28 weeks 29-36 weeks ≥37 weeks	NA	Medication, hospital treatment, ambulatory treatment, and non-medical remedies	German health insurance company	First 3 years of life
Johnston *et al*[35]	1996-1997	Canada (Québec)	Decision-analytic model populated with data from administrative population-based databases	<28 weeks: 1,828 28–32 weeks: 4,685 33–36 weeks: 20,795	<28 weeks 28-32 weeks 33-36 weeks	NA	Direct medical, parental time off work	Régie de l'assurance maladie du Québec (RAMQ) physician billing data from Québec, Canada linked to MED-ÉCHO hospital discharge abstract databases	First ten years of life

Khan *et al*[36]	2009-2010	UK (East Midlands)	Prospective cohort study	32-33 weeks: 85 34-36 weeks: 508 32-36 weeks: 594 ≥37 weeks: 716	32-33 34-36 32-36 ≥37	NA	Neonatal, other hospital, community care, medications, lost earnings, special equipment, adaptations	Bespoke questionnaires designed for The Late and Moderate Preterm Birth Study (LAMBS)	From initial hospital discharge to 24 months
Korvenranta *et al*[38]	2000-2003	Finland	Retrospective national register study	22-23 weeks: 17 24-25 weeks: 135 26-27 weeks: 234 28-29 weeks: 391 30-31 weeks: 771 >31 weeks: 248	<32	<1501	Hospital re-hospitalisations and outpatient visits	Finnish Medical Birth Register and Finnish Hospital Discharge Register	First 3 years of life
Korvenranta *et al*[39]	2001-2002	Finland	Prospective cohort study	<32 weeks: 901 38–42 weeks: 368	<32 38–42	<1501	Emergency outpatient, Nonemergency outpatient, Hospitalisations, Other physician visits, Physical therapy/ occupational therapy, Psychologist, Nurse practitioner, Speech therapist, Other	Finnish Medical Birth Register and Finnish Hospital Discharge Register; parental questionnaires	Fifth year of life

Korvenranta *et al*[40]	2000-2003	Finland	Retrospective national register study	Preterm survivors: 23 weeks: 17 24-25 weeks: 135 26-27 weeks: 231 28-29 weeks: 385 30-31 weeks: 756 Preterm non-survivors: 278 Term: 200,609	<32 37–41	<1501	Nonemergency outpatient visits, emergency outpatient visits, hospital readmissions	Finnish Medical Birth Register and Finnish Hospital Discharge Register	First 4 years of life
Kuzniewicz *et al*[41]	2003-2012	USA (California)	Retrospective cohort study	31-33 weeks: 3696 34-36 weeks: 19,494 37-38 weeks: 74,023 39+ weeks: 212,523	31-33 34-36 37-38 39+	NA	Hospital readmissions, emergency department visits	Kaiser Permanente cohort, integrated health care delivery system, Northern California.	30 days after initial hospital discharge
Luu *et al*[45]	2003-2004	Canada (Quebec)	Prospective cohort study	23–25 weeks : 49 26–28 weeks: 205	23–25 26–28	NA	Inpatient hospital services, outpatient healthcare services, prescribed medication and assistive technology use	Parental questionnaires completed as part of 'The Research Consortium on Children Born Extremely Preterm' cohort, cross-checked against medical charts	From neonatal discharge to 18 months
Mangham *et al*[46]	2006	England and Wales	Decision-analytic model populated with data from administrative population-based databases	≤ 27 weeks: 3185 28-31 weeks: 5575 32-33 weeks: 6410 34-36 weeks: 32,812 ≥37 weeks: 621,618	All	NA	Hospital inpatient, hospital outpatient, community health and social care, education services	Secondary data from external cohorts and cross-sectional studies	First 18 years of life

McLaurin *et al*[47]	2004	USA	Retrospective analysis of insurance database	33-36 weeks: 1683 ≥37 weeks: 33,745	33–36 ≥37	NA	Inpatient hospitalisations, well-infant physician office visits, outpatient hospital services, home health/private nurse, acute care physician office visits, prescription drugs, other professional	MedStat MarketScan Commercial Claims and Encounters database	First year of life
Mourani *et al*[50]	2001-2007	USA	Follow-up of randomised controlled trial	Year 1: 512; years 1-2: 455; years 2-3: 412; years 3-4.5: 282	≤34 weeks with requirement for mechanical ventilation at birth	500-1250	Hospital, outpatient	Prophylactic inhaled nitric oxide trial (iNO) with follow-up through parental interviews and assessment of medical bills	First 4.5 years of life
Petrou *et al*[52]	1995	UK and Republic of Ireland	Prospective cohort study	20-25 weeks: 190, Mainstream school classmates: 141	20-25 Classmates	NA	Hospital inpatient care, outpatient and day care, community health and social care, drugs/medications, education services	Parental questionnaires as part of EPICure whole population cohort study	The 11th year of life
Stephens *et al*[54]	2001-2011	Australia (New South Wales)	Retrospective analysis of regional birth cohort	24-27 weeks: 3659 28-31 weeks: 7235 32-33 weeks: 9007 39-40 weeks: 539,631	24-27 28-31 32-33 39-40	NA	Hospital	Linked New South Wales Perinatal Data Collection, Admitted Patient Data Collection, and Registry of Births, Deaths and Marriages	First 6 years of life

Stephens *et al*[55]	2001-2005	Australia (New South Wales)	Retrospective analysis of regional birth cohort	33-34 weeks: 3878 35-36 weeks: 12,665 37-38 weeks: 80,478 39-40 weeks: 218,254 ≥41 weeks: 77,689	33-34 35-36 37-38 39-40 ≥41	NA	Hospital	Linked New South Wales Perinatal Data Collection, Admitted Patient Data Collection, and Registry of Births, Deaths and Marriages	Between 1 and 6 years of age
Thanh *et al*[56]	2004-2010	Canada (Alberta)	Retrospective analysis of regional birth cohort	<2500g: 16,209 ≥2500g: 189,586	NA	<2500g ≥2500g	Hospital inpatient and outpatient services, practitioner services	Alberta Perinatal Health Program administrative databases	First year of life
Westrupp *et al*[58]	2004-13	Australia	Prospective cohort studies	Birth cohort: Moderate-to-high risk: 297 Mild risk: 442 No perinatal risk: 3,973 Kindergarten cohort: Moderate-to-high risk: 361 Mild risk: 465 No perinatal risk: 3,629	Moderate-to-high risk: <32 Mild risk: 32–36 No perinatal risk: ≥37	Moderate-to-high risk: <1500g Mild risk: 1500–2499g No perinatal risk: ≥2500g	Community-based health care, prescription medications and total Medicare combined costs	Longitudinal Study of Australian Children cohort studies with Medicare data linkage	Up to first 9 years of age
Young *et al*[60]	2000-2010	USA (Utah and Idaho)	Retrospective analysis of insurance database	34-36 weeks: 19,081 37-38 weeks: 94,178 39-42 weeks: 180,144	34-36 37-38 39-42	NA	Hospital	Intermountain Healthcare Enterprise Data Warehouse	Within 28 days of initial discharge

Notes: BPD, bronchopulmonary dysplasia; HO, home oxygen; EPT, extremely preterm; VPT, very preterm; MPT, moderately preterm; MLPT, moderate to late preterm; LMPT, late and moderate preterm; LPT, late preterm; PT, preterm; ET: early term; FT, full term; LT, late term; VLBW, very low birthweight; LBW, low birthweight; NBW, normal birth weight; NA, not applicable.

Appendix 6: Resource use and/or economic costs following the initial hospital discharge

Study	Currency (price date)	Total cost per child including initial hospitalisation (mean)	Total cost per survivor including initial hospitalisation (mean)	Costs following initial hospital discharge (mean) (unless otherwise stated)	Rehospitalisation length of stay (mean days) (unless otherwise stated)
Barradas *et al*[21]	US $ (2015 prices)[±]	Medicaid: 16,200; Commercial: 15,300; Self-pay: 8000	NA	Medicaid: 12,800; Commercial: 12,400; Self-pay: 6400	Medicaid: 7.4; Commercial: 6.8; Self-pay: 4.6
Berard *et al*[22]	Can $ (2003 prices) (1Can$, 2003 prices = 1.04US$, 2015 prices)[#]	NA	NA	LPT: Re-hospitalisation: 1727 (years 0-3), 1530 (years 0-2), 212 (years 2-3). Physician visits: 977 (0-3), 756 (0-2), 221 (2-3). Emergency department visits: 2 (0-3), 1 (0-2), 1 (2-3). Prescriptions: 388 (0-3), 282 (0-2), 106 (2-3) Term: Re-hospitalisation: 628 (years 0-3), 527 (years 0-2), 102 (years 2-3). Physician visits: 766 (0-3), 575 (0-2), 191 (2-3). Emergency department visits: 1 (0-3), 1 (0-2), 1 (2-3). Prescriptions: 257 (0-3), 182 (0-2), 75 (2-3)	NA
Bird *et al*[23]	US $ (2009 prices)[±] (1US$, 2009 prices = 1.10US$, 2015 prices)[#]	NA	Inpatient: 34-36: 3027 37-42: 2183 Outpatient: 34-36: 1560 37-42: 1316 Total health care: 34-36: 4541 37-42: 3472	NA	34-36 (survivors): 0.65 37-42 (survivors): 0.48

Cavallo *et al*[25]	€ (2008 prices) (1€, 2008 prices = 1.45US$, 2015 prices)[#]	NA	NA	≤30 weeks and/or ≤1500g: From discharge to six months: Total public health system cost: 1433 Total societal cost: 13,382 From six months to 18 months: Total public health system cost: 820 Total societal cost: 12,257 Term: From discharge to six months: Total public health system cost: 565 Total societal cost: 12,368 From six months to 18 months: Total public health system cost: 391 Total societal cost: 920	NA
Corchia *et al*[27]	NA	NA	NA	NA	1st year rehospitalisation rate: VPT: 24.8%; MLPT: 16.0%; ET:15.7%; FT: 12.6% 2nd year rehospitalisation rate: VPT: 9.8%; MLPT: 8.4%; ET: 8.9%; FT: 7.5% 3rd year rehospitalisation rate: VPT: 9.7%; MLPT: 5.3%; ET: 5.4%; FT: 4.9%
Gareau *et al*[28]	US $ (2016 prices)	NA	NA	Estimated savings from avoiding poor outcomes: Preterm birth: 22,667 Low birthweight: 29,627	NA
Helle *et al*[30]	€ (2011 prices) (1€, 2011 prices = 1.18US$, 2015 prices)[#]	NA	<32: 45,107* 32-33: 19,872* 34-36: 5621* 37-38: 2987* 39-41: 2700* >41: 2679*	1926* (Year 2); 1116* (Year 3) 995* (Year 2); 517* (Year 3) 870* (Year 2); 551* (Year 3) 788* (Year 2); 482* (Year 3) 742* (Year 2); 452* (Year 3) 748* (Year 2); 457* (Year 3)	NA

Hong *et al*[32]	NA	NA	NA	Mean: No BPD: 1,349, BPD/No-HO: 948, BPD/HO: 897	Amongst survivors: All: 4.42* No BPD: 3.67: BPD/No HO: 6.48 BPD/HO: 8.54
Hummer *et al*[33]	€ (2009 prices) (1€, 2009 prices = 1.35US$, 2015 prices)[#]	NA	NA	<1500g: Outpatient expenditures: 1173.76 (year 0-1); 289.12 (years 2-5); 319.83 (years 6-14); 218.17 (years 15-21) Medical attendance: 207.03 (year 0-1); 208.39 (years 2-5); 256.07 (years 6-14); 176.83 (years 15-21) Medical drugs taken: 966.73 (year 0-1); 80.73 (years 2-5); 63.75 (years 6-14); 41.34 (years 15-21) <2500g: Outpatient expenditures: 320.82 (year 0-1); 233.75 (years 2-5); 219.19 (years 6-14); 202.38 (years 15-21) Medical attendance: 232.56 (year 0-1); 191.98 (years 2-5); 180.64 (years 6-14); 158.86 (years 15-21) Medical drugs taken: 88.25 (year 0-1); 41.77 (years 2-5); 38.55 (years 6-14); 43.52 (years 15-21) ≥2500g: Outpatient expenditures: 227.75 (year 0-1); 198.97 (years 2-5); 194.96 (years 6-14); 184.41 (years 15-21) Medical attendance: 202.37 (year 0-1); 167.89 (years 2-5); 162.94 (years 6-14); 147.56 (years 15-21) Medical drugs taken: 25.38 (year 0-1); 31.09 (years 2-5); 32.01 (years 6-14); 36.85 (years 15-21)	<1500g: 1.21 (years 2-5) 0.72 (years 6-14) 1.19 (years 15-21) <2500g: 0.81 (years 2-5) 0.67 (years 6-14) 0.99 (years 15-21) ≥2500g: 0.54 (years 2-5) 0.44 (years 6-14) 0.73 (years 15-21)

Jacob *et al*[34]	€ (2016 prices)[±]	<28 weeks: 74,009 (year 1) 29-36 weeks: 8565 (year 1) ≥37 weeks: 1590 (year 1)	NA	<28 weeks: 5341 (year 2); 3390 (year 3) 29-36 weeks: 1603 (year 2); 1053 (year 3) ≥37 weeks: 902 (year 2); 756(year 3)	NA
Johnston *et al*[35]	Can $ (2012 prices) (1Can$, 2012 prices = 0.84US$, 2015 prices)[#]	<28 weeks: 67,467 28-32 weeks: 54,554	NA	<28 weeks: 9280 (Medical from discharge to year 2) 1374 (Medical, years 2-4), 875 (Indirect, years 2-4), 1059 (Medical, years 5-10), 782 (Indirect, years 5-10) 28-32 weeks: 6573 (Medical from discharge to year 2) 1936 (Medical, years 2-4), 1136 (Indirect, years 2-4), 1077 (Medical, years 5-10), 912 (Indirect, years 5-10)	<28 weeks: 17.45 (Discharge-year 2) 0.89 (year 2-3) 0.65 (year 3-4) 0.42 (year 4-5) 0.50 (year 5-6) 0.32 (year 6-7) 0.45 (year 7-8) 0.24 (year 8-9) 0.04 (year 9-10) 28-32 weeks: 8.75 (Discharge-year 2) 1.60 (year 2-3) 0.50 (year 3-4) 0.35 (year 4-5) 0.39 (year 5-6) 0.24 (year 6-7) 0.22 (year 7-8) 0.19 (year 8-9) 0.09 (year 9-10)

		33-36 weeks: 10,010		33-36 weeks: 2228 (Medical from discharge to year 2) 1015 (Medical, years 2-4), 805 (Indirect, years 2-4), 814 (Medical, years 5-10), 779 (Indirect, years 5-10)	33-36 weeks: 2.40 (Discharge-year 2) 0.40 (year 2-3) 0.26 (year 3-4) 0.20 (year 4-5) 0.16 (year 5-6) 0.20 (year 6-7) 0.17 (year 7-8) 0.10 (year 8-9) 0.10 (year 9-10)
Khan *et al*[36]	UK£ (2010-11 prices) (1UK£, 2011 prices = 1.51US$, 2015 prices)[#]	32-33 weeks: Neonatal care: 9387.69; Other hospital care: 1411.65; Community care: 960.22; Medications: 13.14; Lost earnings: 252.87; Special equipment: 7.05; Adaptations: 4.24 34-36 weeks: Neonatal care: 2827.01; Other hospital care: 1642.21; Community care: 1059.32; Medications: 7.15; Lost earnings: 255.95; Special equipment: 4.87; Adaptations: 26.97	NA	NA	NA

		32-36 weeks: Neonatal care: 3765.83; Other hospital care: 1069.22; Community care: 1045.14; Medications: 8.01; Lost earnings: 255.51; Special equipment: 5.18; Adaptations: 23.72 ≥37 weeks: Neonatal care: 172.66; Other hospital care; 673.76, Community care: 1005.24; Medications: 7.99; Lost earnings: 155.86 Special equipment: 3.32; Adaptations: 36.70			
Korvenranta *et al*[38]	NA	NA	NA	NA	Rehospitalisation rate (years 1, 2 and 3): 22-23 weeks: 76.5%, 47.1%, 52.9% 24-25 weeks: 65.9, 42.2%, 37.8% 26-27 weeks: 60.7%, 38.5%, 26.1% 28-29 weeks: 53.2%, 30.4%, 19.7% 30-31 weeks: 43.2%, 20.6%, 17.1% >31 weeks: 40.7%, 27.8%, 18.5%

					All: 49.3%, 28.0%, 20.9% Mean outpatient visits (years 1, 2 and 3): 22-23 weeks: 10.5, 9.2, 6.9 24-25 weeks: 10.5, 9.0, 6.0 26-27 weeks: 9.8, 6.7, 4.5 28-29 weeks: 9.7, 5.3, 3.1 30-31 weeks: 7.4, 3.4, 2.0 >31 weeks: 6.8, 4.1, 2.5 All: 8.4, 4.8, 3.0
Korvenranta *et al*[39]	€ (2008 prices) (1€, 2008 prices = 1.23US$, 2015 prices)[#]	NA	NA	VPT with morbidities: 3265 VPT without morbidities: 1023 Term: 749	23 weeks: 3.0; 24-25: 0.3; 26-27: 0.4; 28-29: 0.1; 30-31: 0.2; Term: 0.1
Korvenranta *et al*[40]	€ (2008 prices) (1€, 2008 prices = 1.23US$, 2015 prices)[#]	NA	23 weeks: 175,490 24-25: 143,570 26-27: 105,631 28-29: 72,366 30-31: 45,714 All: 68,073 Term: 4580	Nonemergency outpatient visits (survivors): 23 weeks: 11,156 24-25: 10,657 26-27: 8306 28-29: 7381 30-31: 4797 All: 6411 Term: 720 Emergency outpatient visits (survivors): 23 weeks: 1183 24-25: 1347 26-27: 1367 28-29: 926 30-31: 699 All: 900 Term: 251 Hospital readmissions (survivors): 23 weeks: 15,753 24-25: 10,753 26-27: 7770	23 week (survivors): 14.6 24-25 (survivors): 12.1 26-27 (survivors): 8.6 28-29 (survivors): 8.2 30-31 (survivors): 5.7 All (survivors): 7.2 Term (survivors): 2.0

				28-29: 7661 30-31: 5226 All: 6675 Term: 2275	
Kuzniewicz *et al*[41]	NA	NA	NA	NA	Rehospitalisation rate: 4.2% (31 weeks); 2.9% (32); 2.4% (33); 3.8% (34); 8.4% (35); 9.0% (36); 9.8% (37); 5.4% (38); 3.8% (39); 3.1% (40); 2.5% (41); 1.9% (42) Emergency department visit rate: 6.9% (31 weeks); 4.7% (32); 5.3% (33); 4.7% (34); 4.6% (35); 4.6% (36); 4.2% (37); 4.8% (38); 3.9% (39); 3.9% (40); 4.0% (41); 3.0% (42)
Luu *et al*[45]	NA	NA	NA	NA	Hospital days (including neonatal admission): 23-25 weeks: 124* 26-28 weeks: 88* Long-term rehabilitation care: 23-25 weeks: 29% rate 26-28 weeks: 17% rate Regular prescribed medications: 23-25 weeks: 38% rate 26-28 weeks: 28% rate Any assistive devices: 23-25 weeks: 59% rate 26-28 weeks: 33% rate

Mangham *et al*[46]	UK£ (2006 prices) (1UK£, 2006 prices = 1.72US$, 2015 prices)[#]	NA	Total incremental public sector cost vs term: <28 weeks: 94,740 <33 weeks: 61,781 <37 weeks: 22,885	<28 weeks (incremental cost vs term): Hospital outpatient: 1589; community health and social care: 1621; education: 3978 <33 weeks (incremental cost vs term): Hospital outpatient: 802; community health and social care: 552; education: 1463 <33 weeks (incremental cost vs term): Hospital outpatient: 553; community health and social care: 182; education: 494	NA
McLaurin *et al*[47]	US $ (2008 prices)[±] (1US$, 2008 prices = 1.11US$, 2015 prices)[#]	33-36 weeks: 38,301 ≥37 weeks: 6156	NA	33-36 weeks: 12,247 ≥37 weeks: 4069	33-36 weeks: 4.5 ≥37 weeks: 3.4
Mourani *et al*[50]	US $ (2012 prices) (1US$, 2012 prices = 1.04US$, 2015 prices)[#]	NA	NA	Year 1: Children without readmissions: 9600* Children readmitted only to ward: 30,200* Children with ≥1 ICU readmission: 69,700* Year 2: Children without readmissions: 16,700* Children readmitted only to ward: 36,200* Children with ≥1 ICU readmission: 109,600* Year 3: Children without readmissions: 19,100* Children readmitted only to ward: 43,100* Children with ≥1 ICU readmission: 116,000* Year 4: Children without readmissions: 19,400* Children readmitted only to ward: 53,200* Children with ≥1 ICU readmission: 111,800*	ICU days: Years 0-1: 11.1 Years 1-2: 8.0 Years 2-3: 6.1 Years 3-4.5: 5.7 Days of mechanical ventilation: Years 0-1: 9.3 Years 1-2: 8.3 Years 2-3: 6.6 Years 3-4.5: 5.1
Petrou *et al*[52]	£ (2006-7 prices) (1UK£, 2007 prices = 1.67US$, 2015 prices)[#]	NA	NA	≤25 weeks: Hospital inpatient care: 345.9 Hospital outpatient and day care: 158.0 Community health and social care: 617.0 Drugs/medications: 23.7 Education services: 5339.2	NA

				Total public sector: 6483.9 Classmates: Hospital inpatient care: 48.4 Hospital outpatient and day care: 50.2 Community health and social care: 273.5 Drugs/medications: 11.7 Education services: 3623.0 Total public sector: 4007.3	
Stephens *et al*[54]	US $ (2014 prices) (1US$, 2014 prices = 1.01US$, 2015 prices)[#]	NA	NA	By 2 months of age: 24-27: 26,800 28-31: 9850 32-33: 6890 39-40: 4980	Readmission rates: Within 30 days of initial discharge: 24-27: 30.7% 28-31: 19.9% 32-33: 10.1% 39-40: 3.4% Year 0-1: 24-27: 62.1% 28-31: 47.7% 32-33: 36.2% 39-40: 16.8% Year 1-2: 24-27: 36.0% 28-31: 25.3% 32-33: 21.1% 39-40: 13.3% Year 2-3: 24-27: 28.4% 28-31: 18.3% 32-33: 16.0% 39-40: 9.8% Year 3-4: 24-27: 19.3% 28-31: 15.7% 32-33: 13.2% 39-40: 8.7% Year 4-5:

					24-27: 16.8% 28-31: 13.3% 32-33: 11.5% 39-40: 8.1% Year 5-6: 24-27: 14.8% 28-31: 11.0% 32-33: 10.0% 39-40: 7.0%
Stephens *et al*[55]	Aus $ (2013 prices) (1Aus$, 2013 prices = 0.69US$, 2015 prices)[#]	NA	NA	Cost per readmission: 33-34 weeks: 2645 35-36 weeks: 2651 37–38 weeks: 2610 39–40 weeks: 2591 ≥41 weeks: 2576	Odds of readmission: 33-34 weeks: 1.39 35-36 weeks: 1.19 37–38 weeks: 1.10 39–40 weeks: 1.00 (Referent) ≥41 weeks: 0.98
Thanh *et al*[56]	Can $ (2013 prices) (1Can$, 2013 prices = 0.83US$, 2015 prices)[#]	<2,500g: 33,096 ≥2,500g: 3,942	NA	Inpatient services: <2500g: 11,834 ≥2500g: 1161 Outpatient visits: <2500g: 1297 ≥2500g: 424 Practitioner services: <2500g: 1858 ≥2500g: 701	Hospital days: <2,500g: 7.4 ≥2,500g: 0.4 NICU days: <2,500g: 6.0 ≥2,500g: 0.1
Westrupp *et al*[58]	Aus $ (2011 prices) (1Aus$, 2011 prices = 0.70US$, 2015 prices)[#]	NA	NA	Birth cohort (first six years): Moderate-to-high risk: - Community health care: 2242 - Prescribed medications: 162 - Total Medicare: 2405 Mild risk: - Community health care: 2284 - Prescribed medications: 197 - Total Medicare: 2481 No perinatal risk: - Community health care: 1977 - Prescribed medications: 111	NA

				- Total Medicare: 2088 Kindergarten cohort (first six years): Moderate-to-high risk: - Community health care: 1468 - Prescribed medications: 257 - Total Medicare: 1725 Mild risk: - Community health care: 1366 - Prescribed medications: 179 - Total Medicare: 1545 No perinatal risk: - Community health care: 1208 - Prescribed medications: 111 - Total Medicare: 1319	
Young *et al*[60]	NA	NA	NA	NA	Readmission rate per 1,000 newborns: 34-36 weeks: 34.6 37-38 weeks: 20.6 39-42 weeks: 14.8

Notes: OHs, other hospitalisations; GPC, General Practitioner consultation; SVs, specialist visits; OSVs, other specialist visits; LATs, laboratory tests; ITs, imaging tests; RT, rehabilitation therapy; PS, psychological support; HAs, hospital admissions; PT/OT, Physical Therapy/Occupational Therapy; EAD, early Discharge; LAD, late Discharge; ICU, Intensive Care Unit; NICU, Neonatal Intensive Care Unit; BPD, bronchopulmonary dysplasia; HO, home oxygen; LOS, length of stay; EPT, extremely preterm; VPT, very preterm; MPT, moderately preterm; MLPT, moderate to late preterm; LMPT, late and moderate preterm; LPT, late preterm; PT, preterm; ET, early term; FT, full term; VLBW, very low birthweight; LBW, low birthweight; NBW, normal birth weight; NA, not applicable; BPD, bronchopulmonary dysplasia; HO, home oxygen.

[±] As currency price dates not reported, it was assumed that the costs used in the valuation process applied to the financial year prior to study publication.

[#] Cost conversion rates reflecting relevant country-specific Gross Domestic Product deflator indices and purchasing power parities supplied by the OECD. These rates vary across individual countries using the same currency such as the €.[17] * Number expressed as median.