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The long-term costs of preterm birth and low birth weight: results of a systematic review

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Abstract

The high rates of morbidity and mortality arising from preterm birth and low birth weight impose an immense burden on the health, education and social services, and on families. This paper presents the results of a systematic review of the published and unpublished evidence regarding the long-term economic implications of preterm birth and low birth weight for various sectors of the economy and for individuals. The paper highlights the variable methodological quality of the bulk of long-term economic studies of preterm birth and low birth weight and suggests ways in which these methodological limitations can be overcome. The paper reveals that preterm birth and low birth weight can result in substantial costs to the health sector following the infant's initial discharge from hospital. It can also impose a substantial burden on special education and social services, on families and carers of the infants and on society generally. In addition to the costs identified by the literature, preterm birth and low birth weight can have other long-term consequences that require evaluation from an economic perspective.

Keywords: systematic review, costs, long-term, preterm birth, low birth weight

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Introduction

The high rates of morbidity and mortality arising from preterm birth and low birth weight impose an immense burden on the health, education and social

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services, and on families. This was evaluated in a pivotal paper published in the *New England Journal of Medicine* in the early 1980s (Boyle *et al*. 1983), which found that for infants of < 1500 g admitted to neonatal intensive care units, the long-term costs outweighed the measurable economic benefits. Since the publication of that study, the incidence of preterm birth and low birth weight has increased. For example, the incidence of live-born babies weighing < 1500 g in England and Wales increased from 0.84% in 1983 to 1.22% in 1998 and that of live-born babies weighing < 1000 g from 0.27% to 0.48% (Macfarlane & Mugford 2000). Similar trends have been noted in other industrialized nations (OECD 1999). Furthermore, advances in perinatal practices, such as increased use of assisted ventilation in the delivery room and surfactant therapy, have improved the chances for survival of extremely low gestational age and birth weight infants. These epidemiological trends are likely to increase the demand for and cost of specialist services for preterm and low birth weight infants, requiring strategic co-ordination of services between agencies.

There have been numerous studies of the costs of preterm birth and low birth weight incurred during the neonatal stay (Petrou *et al*., in press), but less is known about their long-term economic impact. This paper will systematically review published and unpublished evidence regarding the economic implications of preterm birth and low birth weight following the discharge of the infant from the neonatal unit. It will also summarize the scope and scientific quality of the published and unpublished evidence in order to identify gaps in our knowledge and to consider the future research agenda in this area. The paper is based on research conducted by one of the authors (TS) as part of a master's degree.

Methods

Several strategies were combined to identify relevant studies. First, a number of computerized databases were searched: MEDLINE, CINAHL, EconLit, Science Citation Index (SCI), Social Science Citation Index, Index to Scientific and Technical Proceedings (ISTP), British Library Inside Information (BLII), EMBASE, Cochrane Library (CDSR), York Database of Abstracts of Reviews of Effectiveness (DARE), NHS Economic Evaluation Database (NEED) and the Database of Consortium of University Research Libraries (COPAC). Second, formal searches of the ASLIB Index to British Theses and Current Research in Britain (CRIB) were conducted for masters and PhD theses accepted by British universities addressing economic aspects of preterm

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birth and low birth weight. Third, books and pamphlets held in the National Perinatal Epidemiology Unit library with material relating to economic aspects of preterm birth and low birth weight were hand searched to determine their relevance to the study. Fourth, published and unpublished manuscripts identified through fellow health economic researchers and by other means were reviewed to determine their relevance to the study. Fifth, bibliographies of economic evaluations (Backhouse, Backhouse & Edey 1992; Elixhauser *et al*. 1993) were scanned for studies that might have been missed using other sources.

The search terms applied to all electronic databases were developed and tested on MEDLINE using the MeSH (Medical Subject Headings) thesaurus and covered all topics contained within the MeSH terms 'prematurity', 'preterm birth' and 'low birth weight'. Pilot searches led us to exclude the MeSH terms 'newborn', 'neonatal' and 'infant' from our search strategy as they proved to be too broad and resulted in a large quantity of irrelevant material, including many economic studies of infants born at term or at normal birth weight. The reference lists of all papers identified by the searches were scrutinised for additional studies. The years 1980–1999 formed the timeframe for the systematic review, as the results of earlier studies were not considered to be generalisable to the present health care context. Furthermore, studies were excluded from the literature searches if they had been conducted in developing countries, if the abstract had not been published in the English language or if the focus was animal research.

The title and abstract of each study identified by the literature searches were considered by the investigators in order to select studies that appeared to report relevant economic research. The report of each of the selected studies was considered by the same investigators and accepted for inclusion in this review if it explicitly described, measured and valued the economic implications of care provided to preterm (< 37 weeks gestation) or low birth weight (< 2500 g) infants following their discharge from the neonatal unit. At both stages in the selection process, disagreements as to whether studies met the investigators' criteria were resolved by discussion. Twenty studies met the criteria for inclusion in the review, out of 1023 initially identified by the literature searches. Six of these 20 studies were economic evaluations that were designed to measure the costs and consequences of interventions targeted at preterm birth or low birth weight, whilst 14 were cost studies. Further details are given in Fig. 1.

The methodological robustness of the 20 selected studies was assessed using guidelines developed by a group of leading health economists and published by the *British Medical Journal* (Drummond & Jefferson 1996). The study design, data collection methods and analysis and interpretation of the results

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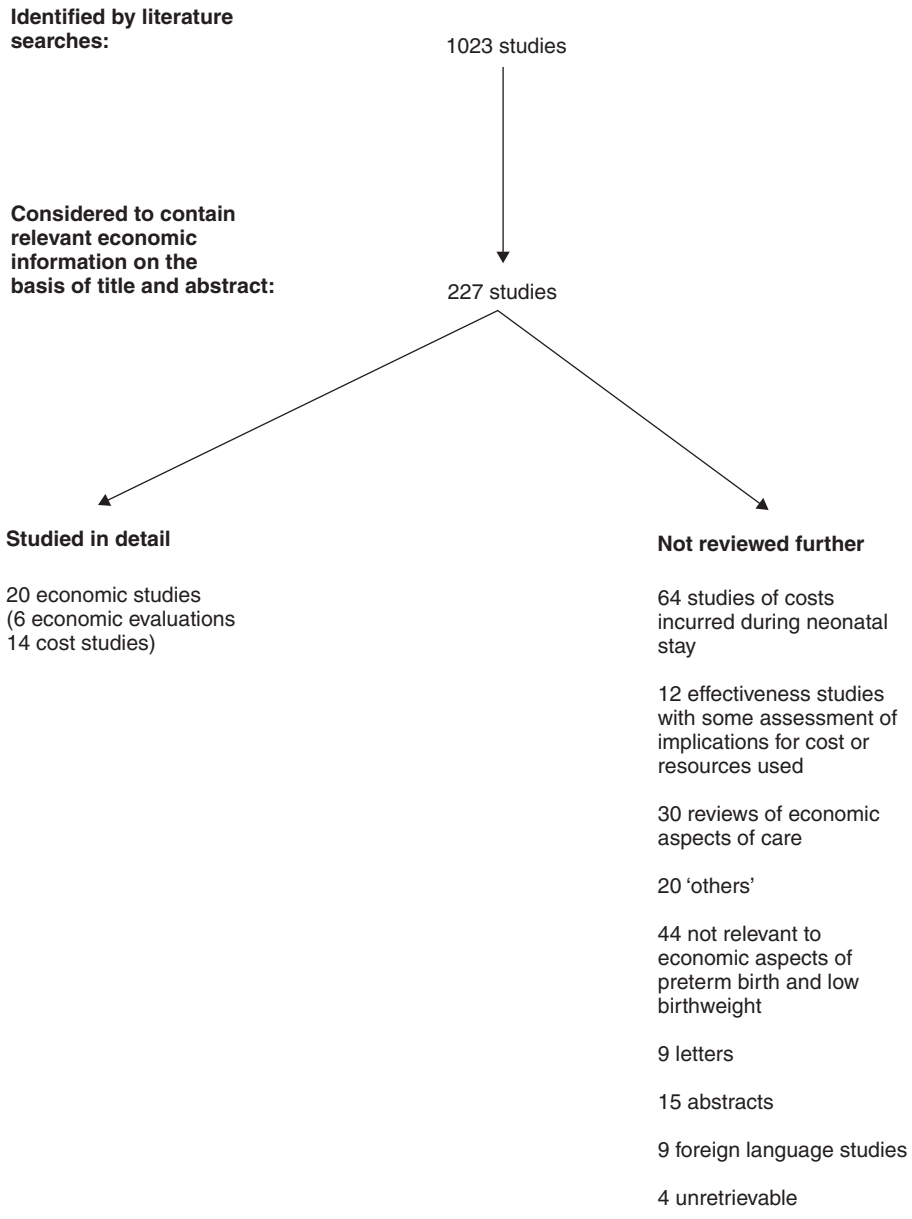

Figure 1 A schematic overview of the results of the literature searches.

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of the economic evaluations were independently assessed by the investigators using the complete checklist of 35 items contained within these guidelines, whilst the cost studies were independently assessed using a subset of 16 applicable items drawn from this checklist. Disagreements as to whether the studies met the requirements of the guidelines were resolved by discussion between the investigators.

All cost data were converted from their respective currencies into UK pounds sterling using Purchasing Power Parities supplied by the Organization for Economic Co-operation and Development. Once converted to UK pounds sterling, the cost data were inflated to 1998 prices using the NHS Hospital and Community Health Services Pay and Prices Inflation Index. Methodological variations between studies prevented a pooling of economic data akin to the meta-analyses performed on the clinical effectiveness literature. Rather, the results of the studies are presented and discussed in a qualitative manner according to the study perspective.

Results

Table 1 summarises the population studied, intervention or condition evaluated, cost categories considered, methodological limitations and principal findings of each of the 20 economic studies included in the review. The remainder of this section discusses the methods used by these studies and their results, which are reported by study perspective.

Methods used by studies

A number of methodological issues were identified by the guidelines used to assess each economic study (Drummond & Jefferson 1996). Three studies in this systematic review estimated the economic implications of preterm birth and low birth weight within the context of randomised controlled trials (RCTs) (Brooten *et al.* 1986; Goetze *et al.* 1993; Backhouse *et al.* 1994). Although RCTs confer the advantage of minimising bias in comparisons between forms of care, the clinical trial protocol itself may induce additional resource use. Therefore, these three studies may have overestimated the costs that would have been incurred in a routine care environment. One study from Canada (Boyle *et al.* 1983), one study from the United States (Walker *et al.* 1984) and two studies from Australia (Kitchen *et al.* 1993; Victorian Infant Collaborative Study Group 1997) estimated the long-term costs and outcomes of population based cohorts

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Table 1 Methodology and results of studies of long-term costs of preterm birth and low birth weight

Author(s)	Population studied	Intervention/condition evaluated	Cost categories considered	Methodological limitations	Principal findings
Economic evaluations					
Boyle *et al.* (1983)	All infants with birth weights of < 1500 g born in Hamilton-Wentworth County in Canada during the years 1964–1979 and 1973–1977.	Regionalised neonatal intensive care.	Health, social services and education services between birth and predicted time of death.	A number of resource use and unit cost estimates were derived from secondary sources.	Long-term costs outweighed the measurable economic benefits.
Walker *et al.* (1984)	247 infants born at < 1000 g in the state of Rhode Island in the US.	Neonatal intensive care.	Health, social services and education services between birth and predicted time of birth.	Charges were not adjusted to true costs. Sensitivity analysis not performed.	Long-term charges outweighed the measureable economic benefits.
Walker *et al.* (1985)	All infants with birth weights < 1500 g born in the state of Rhode Island in the US during the years 1974–1975 and 1979–1980.	Regionalised neonatal intensive care.	Health and education services between birth and predicted time of death.	Charges were not adjusted to approximate to true costs. Sensitivity analysis not performed.	Long-term charges outweighed the by measurable economic benefits.
Javitt *et al.* (1993)	Hypothetical cohorts of preterm infants with birth weights of 500–749 g, 750–999 g and 1000–1249 g.	Cryotherapy for treatment of retinopathy of prematurity.	Health, social services, education and other services and lost productivity throughout infancy and adulthood.	No primary costing research was conducted. Charges were not adjusted to approximate to true costs.	Appropriately timed screening for and treatment of retinopathy of prematurity predicted to result in net budgetary savings.
Backhouse *et al.* (1994)	1237 infants in North America with birth weights of ≥ 1250 g and suffering from respiratory distress syndrome.	Synthetic surfactant administered to infants receiving mechanical ventilation.	Health services consumed during the first year of life.	No primary costing research was conducted. Charges were not adjusted to approximate to true costs.	Hospital charges for infants treated with surfactant were less than those for a comparable cohort receiving air placebo.

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Table 1 (*continued*)

Author(s)	Population studied	Intervention/condition evaluated	Cost categories considered	Methodological limitations	Principal findings
Victorian Infant Collaborative Study Group (1997)	1340 infants born in Victoria, Australia, with birth weights of 500–999 g.	Neonatal intensive care during three distinct eras: (1) 1979–1980; (2) 1985–1987; and (3) 1991–1992.	Health services consumed during first 2 years of life.	No primary costing research was conducted.	The incremental cost of improving the outcomes of extremely low birth weight infants has fallen in the post surfactant era.
Cost studies					
Papiernik (1983)	Hypothetical cohort of low birth weight twins.	Economic implications of multiple births.	Consumption of health services. Time frame for analysis not specified.	No primary costing research was conducted. Sensitivity analysis not performed.	Low birth weight twins estimated to cost up to 10 times more than a singleton born at term.
Brooten *et al.* (1986)	79 infants born in Pennsylvania, US, at < 1500 g.	Early hospital discharge and home follow-up when infants satisfied a standard set of criteria.	Health services consumed during first 18 months of life.	Charges were not adjusted to approximate to true costs. Sensitivity analysis not performed.	A policy of early hospital discharge, with follow-up care in the home, resulted in substantial cost savings.
Pharoah *et al.* (1988)	152 infants born at < 1500 g in Merseyside, UK.	Care provided to infants following their discharge from neonatal intensive care units.	Health services consumed during the first 4 years of life and lifetime consumption of education, social and other services.	Estimation of education and social service costs not based on primary research. Sensitivity analysis not performed.	After the age of 4, the cost of special education and residential care dominated total care costs.
Papiernik & Keith (1990)	Hypothetical cohort of twin pregnancies at risk of delivering preterm.	Early diagnosis through ultrasound screening and an extra 11 weeks of work leave to expectant mothers.	Consumption of health services and lost productivity. Time frame for analysis was not specified.	No primary costing research was conducted. Discounting and sensitivity analysis not performed.	The preventive intervention was estimated to lead to a two-thirds reduction in care costs.

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Table 1 *(continued)*

Author(s)	Population studied	Intervention/condition evaluated	Cost categories considered	Methodological limitations	Principal findings
Chaikind & Corman (1991)	Approximately 8000 children (aged 6–15 years) born in the US at < 2500 g.	Care provided to the children born at low birth weight.	Special education services provided between the ages of 6 and 15 years.	No primary costing research was conducted. Sensitivity analysis not performed.	Low birth weight resulted in an estimated incremental cost to the US education service of $322.9 million per annum.
McCormick *et al.* (1991)	32 infants born at < 1500 g in Philadelphia, US.	Care provided to infants following their discharge from neonatal intensive care units.	Consumption of health services and parental out of pocket expenses during the first year of life.	Charges were not adjusted to approximate to true costs. Sensitivity analysis not performed.	Costs attributed to very low birth weight infants were approximately nine times those attributed to term infants.
Stevenson *et al.* (1991)	109 infants born at < 1500 g in Merseyside, UK.	Care provided to infants following their discharge from neonatal intensive care units.	Health services consumed during first 4 years of life and lifetime consumption of education and social services.	Estimation of education and social service costs not based on primary research. Sensitivity analysis not performed.	Lifetime care costs diminished substantially with increased birth weight.
Goetze *et al.* (1993)	52 infants born at < 1500 g and diagnosed with moderate to severe bronchopulmonary dysplasia and a need for home oxygen therapy.	Two intensitites of service: (1) high intensity early intervention programme; or (2) low intensity traditional hospital follow-up.	Health services consumed during first 2 years of life.	Discounting and sensitivity analysis not performed.	Annual costs of the high intensity group were approximately triple the cost of the low intensity group.
McCallum & Turbeville (1994)	50 preterm infants born at < 1500 g with intraventricular haemorrhage.	The administration of ventriculo-peritoneal shunts.	Health services consumed during first 20 years of life.	Discounting and sensitivity analysis not performed. Price date not stated.	Care costs for these infants exceeded £3000 per annum after the initial hospitalisation.

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Table 1 *(continued)*

Author(s)	Population studied	Intervention/condition evaluated	Cost categories considered	Methodological limitations	Principal findings
Lewit *et al.* (1995)	All infants and children born in the US at < 2500 g.	Care provided during the calendar year 1988.	Consumption of health, education and social services during 1 calendar year.	Sensitivity analysis not performed.	Incremental cost of low birth weight estimated to cost the US economy between £5.5 and £6 billion in 1988.
Gennaro (1996)	224 preterm infants in Pennsylvania, US, 118 of whom were born at between 1510 g and 2550 g, and 106 of whom were born at < 1500 g.	Care provided to infants following their discharge from one neonatal intensive care unit.	Out of pocket expenses and lost productivity during the first 6 months of life.	Sensitivity analysis not performed.	Out of pocket expenses accounted for between 2% and 4% of the families' annual income.
Stevenson *et al.* (1996a)	399 infants born at < 1500 g and 547 infants born at 1500–2000 g, without clinical disability in Merseyside, UK.	Care provided to infants following their discharge from neonatal intensive care units.	Consumption of hospital and and family practitioner health services during the first 8–9 years of life.	Sensitivity analysis not performed.	Low birth weight children consumed services more intensively throughout the follow-up period than a matched group of controls.
Stevenson *et al.* (1996b)	52 low birth weight infants with clinical disability in Merseyside, UK.	Care provided to infants following their discharge from neonatal intensive care units.	Consumption of health and education services during the first 8–9 years of life.	Sensitivity analysis not performed.	Special education costs accounted for slightly more than half of total care costs for this cohort.
Rogowski (1998)	All live singletons born at < 1500 g in California, US, during the years 1986–1987, that were continuously eligible for the state's Medicaid programme.	Care provided to infants following their discharge from neonatal intensive care units.	Consumption of health services during the first year of life.	Cost estimated assumed to reflect adjusted charges. Sensitivity analysis not performed.	Average care costs increase substantially as birth weight diminishes.

before and after access to specific aspects of neonatal intensive care were available. Two studies (McCormick *et al.* 1991; Stevenson *et al.* 1996a) derived cost estimates for geographically defined cohorts matched to a group of contemporaneous controls. The remaining 11 studies were based on resources consumed by unmatched case series of preterm or low birth weight infants.

No studies used primary research alone to derive resource use and unit cost data, all making some use of existing accounting, administrative and clinical records. Two economic evaluations (Javitt, Dei Cas & Chiang 1993; Victorian Infant Collaborative Study Group 1997) and three cost studies (Papiernik 1983; Papiernik & Keith 1990; Chaikind & Corman 1991) used secondary sources alone to derive cost estimates and, as a result, arrived at conclusions that may not be generalisable to the populations studied. The remaining studies used a hybrid of primary and secondary research methods to derive resource use and unit cost data.

Four economic evaluations (Walker *et al.* 1984; Walker, Vohr & Oh 1985; Javitt, Dei Cas & Chiang 1993; Backhouse *et al.* 1994) and three cost studies (Brooten *et al.* 1986; McCormick *et al.* 1991; Rogowski 1998) cite charges for care rather than costs and may therefore have included elements arising from corporate financial decisions (Finkler 1982). All of these studies were conducted in the United States where there is a comprehensive system of billing and fee-for-service payment of providers. Only one of these studies (Rogowski 1998) adjusted the charges to approximate more closely to the actual costs of resource inputs. The remaining studies used alternative approaches based on cost accounting methods, either using detailed information about the resources used by individual patients (the 'bottom up' approach), or by allocation of total costs by organizational workload (the 'top down' approach).

The majority of the studies considering costs beyond the initial hospital stay considered the long-term economic implications of preterm birth and low birth weight to health providers (17 in total). In addition, nine studies (Boyle *et al.* 1983; Walker *et al.* 1984; Walker, Vohr & Oh 1985; Pharoah *et al.* 1988; Chaikind & Corman 1991; Stevenson *et al.* 1991; Javitt, Dei Cas & Chiang 1993; Lewit *et al.* 1995; Stevenson *et al.* 1996b) estimated the long-term economic implications of preterm birth and low birth weight to education authorities; six (Boyle *et al.* 1983; Walker *et al.* 1984; Pharoah *et al.* 1988; Stevenson *et al.* 1991; Javitt, Dei Cas & Chiang 1993; Lewit *et al.* 1995) to social services; and two (Pharoah *et al.* 1988; Javitt, Dei Cas & Chiang 1993) to other sectors of the economy. Two studies (McCormick *et al.* 1991; Gennaro 1996) estimated out of pocket expenses, such as additional child care costs and hospital visiting costs; and three studies (Papiernik & Keith 1990; Javitt, Dei Cas &

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Chiang 1993; Gennaro 1996) valued the lost productivity that results from the child's illness.

Furthermore, three of the cost studies (Papiernik & Keith 1990; Goetze *et al.* 1993; McCallum & Turbeville 1994) failed to discount future care costs to present values. Discounting is a process normally used by economists to weight current resources more highly than future resources. This failure to discount results in an overestimation of the future costs of preterm birth and low birth weight. Consequently, the results of these three studies must be recognized as an overestimation of the true care costs.

The important tool of sensitivity analysis was only applied by four of the 20 economic studies (Boyle *et al.* 1983; Javitt, Dei Cas & Chiang 1993; Backhouse *et al.* 1994; Victorian Infant Collaborative Study Group 1997). Sensitivity analysis is an approach commonly used by health economists to explore the implications of uncertainty and is used to test the statistical properties of economic parameters. The failure to analyse the uncertainty surrounding key economic parameters leaves the reader unable to judge the degree to which the conclusions of these studies are meaningful and robust (Briggs & Sculpher 1995).

The economic evaluations met an average of 73% of applicable items on the *British Medical Journal* checklist used to assess methodological robustness (range: 62%–83%). The cost studies, on the other hand, met an average of only 54% of applicable items on the checklist (range: 18%–82%). There was no evidence that the methodological robustness of either the economic evaluations or cost studies varied by date of publication. The subsequent discussion of the study results should only be considered in the light of the above methodological issues.

Results of studies

Health care

A number of economic studies of preterm or low birth weight infants provide detailed information about the utilization of health care services following their discharge from the neonatal unit. Each of the studies found that preterm or low birth weight infants are significantly more likely to be rehospitalized than infants born at full term or at normal birth weight. Brooten *et al.* (1986), for example, examined rehospitalizations and acute care visits among 79 very low birth weight infants (≤ 1500 g) during the 18-month period following discharge from the neonatal intensive care unit. They found that 20 infants (25%) had

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to be rehospitalized, mainly as a result of respiratory problems such as pneumonia, and 65 infants (82%) required acute care visits. Furthermore, the increased propensity to consume health care services following discharge from the neonatal unit is not restricted to hospital services. McCormick *et al.* (1991), for example, recorded the utilization of community health care services made during the first year of life by 32 infants born at < 1500 g. The investigators found that these very low birth weight infants made twice as many family practice visits during the first year of life than a matched group of infants born at full term and selected from the same clinical centre.

The studies that have attached a monetary value to the additional health care resources consumed by preterm or low birth weight infants following their discharge from the neonatal unit are of varying methodological quality and differ with regard to the nature of their comparison and control groups, duration of follow-up, and the measurement and classification of outcomes. A study by Rogowski (1998), based on all very low birth weight single live births (< 1500 g) in the state of California Medicaid programme between 1986 and 1987, estimated first year medical costs at ≈ £63 300, £56 900 (89.9%) of which was accounted for by the initial hospitalization. This compares to an average of £1200 in the initial hospitalization and £2600 in first year medical costs for all US births in 1989 (Rogowski 1998). Thus, there is a 47-fold differential in initial hospitalization costs between babies born < 1500 g in the California Medicaid programme and all US births. The differential in first year medical costs between very low birth weight infants and all infants in the United States, by contrast, is much lower at 24-fold. The declining differential in medical costs between the two groups is largely explained by high mortality rates for very low birth weight infants. McCormick *et al.* (1991) also attached a monetary value to the health care services consumed by their study sample following their discharge from the neonatal intensive care unit. Using information collated through parent-completed monthly diaries and quarterly telephone interviews, they estimated first year health care costs at £7748 per very low birth weight infant. This compared to an average cost of £901 for their comparative group of full term infants.

The increased use and cost of health care services consumed by preterm or low birth weight infants persists into childhood. The major neurological abnormalities experienced by these infants, such as cerbral palsy, unilateral or bilateral blindness, deafness requiring hearing aids, and subnormal cognitive function result in significant increases in the use and cost of hospital and family practitioner services over the longer term. This is best illustrated by a series of studies conducted by researchers in Merseyside, England (Pharoah *et al.* 1988;

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Stevenson *et al.* 1996a,b). The investigators followed up a population-based cohort of low birth weight infants (≤ 2000 g) born in 1979–1981 and recorded their use of hospital and family practitioner services up to age 8–9 years. Amongst infants with disability, mean health service costs for the entire 8–9 years follow-up period were estimated at £14 510 for the lowest birth weight group (≤ 1000 g), £12 051 for the intermediate birth weight group (1001–1500 g) and £7178 for the highest birth weight group (1501–2000 g). Amongst infants without disability, it was found that the low birth weight children (≤ 2000 g) used hospital and family practitioner services more intensively throughout the follow up period than a group of controls, matched for age, sex and school class. Indeed, there was a fivefold differential in mean health service costs per child between low birth weight infants without disability and the control infants they were matched to. This differential increased to 16-fold amongst the lowest birth weight group (< 1000 g).

Education

Although the majority of survivors of low gestational age and birth weight are ambulatory and attend school, they experience high rates of school failure and learning problems. Several studies have considered the economic implications of additional education assistance required as a result of these adverse outcomes (Boyle *et al.* 1983; Walker *et al.* 1984; Walker, Vohr & Oh 1985; Pharoah *et al.* 1988; Chaikind & Corman 1991; Stevenson *et al.* 1991, 1996b; Javitt, Dei Cas & Chiang 1993; Lewit *et al.* 1995). One such study investigated the relationship between low birth weight, enrolment in special education and special education costs in the United States (Chaikind & Corman 1991). Using a sample of ≈ 8000 children aged 6–15 who were in school, they calculated the probability of a child attending special education. Children who weighed < 2500 g at birth were almost 50% more likely to be enrolled in any type of special education than children who were of normal weight at birth. This resulted in an estimated incremental cost to the United States education services of £322.9 million per year due to low birth weight.

Stevenson *et al.* (1996b) estimated the cost of special education services among their cohort of low birth weight infants followed up in Merseyside. They calculated that, amongst children with disability, special education costs incurred by 8–9 years of age were greater than the total cost of health services used throughout the follow-up period, including acute health care costs incurred during the initial hospitalization. A cost per disabled child of £42 102 was calculated during those early years of education.

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In addition to increased use of special education services, Lewit *et al.* (1995) report an increased risk of grade repetition and eventual school drop-out among preterm or low birth weight infants. Although no attempt is made to quantify the long-term consequences, it is inferred that this may lead to a lower earnings potential, an increased propensity to commit crime and an increased reliance on social welfare.

Social Services

The high neurosensory and cognitive disability rates among preterm or low birth weight infants also have economic implications for the social services. The parents of these infants often require support from social service departments during the immediate period following the infant's discharge from the neonatal unit. In later life, developmental services–which include day care programmes, case management and counselling, respite care and residential care–may also be required to supplement health and educational services. The cost of these services have been estimated, in part, by studies conducted in the United States and Great Britain. The studies by Boyle *et al.* (1983), Stevenson *et al.* (1991), Javitt, Dei Cas and Chiang (1993) and Lewit *et al.* (1995) do not present disaggregated social service costs within their total cost estimates. However, cost estimates of the lifetime residential needs of preterm and low birth weight infants have been made by two studies. Walker *et al.* (1984) estimated discounted lifetime residential costs for a low birth weight infant at £87 770. This compared to a discounted lifetime residential cost of £520 582, calculated in a British setting (Pharoah *et al.* 1988). Both cost estimates were calculated in the early 1980s and were based on very limited information about the life expectancy and long-term disability of low gestational age and birth weight survivors, and about likely lifetime care needs. Furthermore, both studies made restrictive assumptions about the availability and duration of residential care for adults born at low gestational age and birth weight.

Out of pocket expenses

The health, education and social consequences of preterm birth and low birth weight have economic implications for the families and informal carers of the infants. These costs originate in the additional travel to health and social care providers, child care arrangements for siblings and accommodation expenses which result from the care process. McCormick *et al.* (1991) used diaries and quarterly interviews to estimate the costs incurred by parents during the first year following the infant's discharge from the neonatal intensive care unit.

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Travel costs were estimated at US$15 per month, child care costs at US$47 per month and other expenses at US$62 per month (price date not reported). Travel costs and other expenses were significantly higher than those incurred by parents of a matched group of control infants born at full term. Another American study (Gennaro 1996) interviewed 224 families of low birth weight infants and estimated that out of pocket expenses, attributable to the infant's illness, accounted for between 2% and 4% of the families' annual income.

Wider costs to society

Research on the wider costs of preterm birth and low birth weight is limited and is focused on the first year of life. The most interesting findings relate to the impact of preterm birth and low birth weight on the employment behaviour of the parents. Many mothers of preterm or low birth weight infants who had intended to return to work after the birth either postponed doing so, reduced their hours or left the workforce altogether to care for their child. This was usually associated with a reduction in family income; a 32% reduction is cited in one paper (Gennaro 1996). Javitt, Dei Cas and Chiang (1993) considered the wider budgetary impact of retinopathy of prematurity. Using modelling techniques, they estimated that during adulthood, each case of retinopathy of prematurity will cost central government £4163 per year in social security and disability payments, and £2187 per year in lost tax revenue. The studies by Papiernik and Kieth (1990), Javitt, Dei Cas and Chiang (1993) and Gennaro (1996) described, but did not estimate, some of the intangible costs associated with caring for preterm or low birth weight infants. These include the emotional and physical energy required to care for the infant and the consequent isolation and restricted social contact that ensues.

Discussion

This systematic review of the literature has revealed that preterm birth and low birth weight can result in substantial costs to the health sector following the infant's initial discharge from hospital, even among non-disabled survivors. They can also impose a substantial burden on special education and social services, on the families and carers of the infants, and on society generally.

Many of the studies included in the review were conducted in the United States. The generalisability of the results of these studies to the British setting are limited by differences in the coverage of the health and social care systems, as well as the greater availability of public transportation and shorter distances

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in the UK than would be typical of much of the United States. Nevertheless, these results suggest that decision-makers should be aware of the substantial long-term economic impact of preterm birth and low birth weight and should be sensitive to the financial constraints faced by parents on low incomes at a critical time in the parent–child relationship.

In addition to the costs identified by the literature, preterm birth and low birth weight can have other long-term consequences that require evaluation from an economic perspective. Institutionalization costs for physically handicapped and mentally retarded infants have not been widely reported despite continued institutionalization practices in many industrialized nations (Waitzman, Romano & Scheffler 1994). The use of day care services and respite care has similarly been overlooked by economic studies of preterm birth and low birth weight. Other costs that have been overlooked include costs borne by local authorities and voluntary organizations–such as adaptations that have to be made to the infant'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 particular, no empirical research has estimated the economic implications of preterm birth and low birth weight to families and informal carers beyond the first year of life. In addition to the costs of travel, child care and accommodation, other potential costs faced by families and informal carers 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. Furthermore, no monetary valuation of the intangible consequences of preterm birth and low birth weight, such as the pain, fear, suffering and emotional and social isolation experienced by the infants themselves and their carers, has been attempted. The relative paucity of estimates of the long-term costs of preterm birth and low birth weight can partly be explained by a lack of routinely collected epidemiological data in this area. However, as the survival profile for preterm and low birth weight infants improves and further information on developmental outcomes during adolescence and adulthood becomes available, opportunities for economic research will increase.

The review highlighted the variable methodological quality of the bulk of long-term economic studies of preterm birth and low birth weight. Detailed and disaggregated information on resource use and unit costs was commonly not provided. The important tools of discounting and sensitivity analysis were often not used at all or only used in part. The economic evaluations were more successful than the cost studies in meeting the methodological requirements of the checklist produced by the British Medical Journal Economic Evaluation

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Working Party (Drummond & Jefferson 1996). This may be the result of an increased tendency for economic evaluations to be conducted by health economists, rather than health service researchers or clinicians. As checklists and guidelines for economic evaluation are more widely disseminated, economic studies of preterm birth and low birth weight will require a more consistent application of robust methodological standards.

Estimates of the long-term costs of preterm birth and low birth weight can be informative for decision-makers. For example, cost estimates for each sector of the economy can inform the planning of services. Broad economic aggregates, such as the lifetime costs attributable to the specific sequelae of preterm birth and low birth weight, can provide a basis for assessing competing strategies for research and prevention. However, it should be noted that cost data alone cannot identify the most efficient allocation of finite health care resources. Rather, it is information on incremental costs and incremental health gains attributable to particular health care activities that can identify the combination of human and material inputs that maximise health benefits. This can be achieved through the general framework of economic evaluation. Only six of the studies included in the systematic review were economic evaluations (Boyle *et al.* 1983; Backhouse *et al.* 1994; Walker *et al.* 1984; Walker, Vohr & Oh 1985; Javitt, Dei Cas & Chiang 1993; Victorian Infant Collaborative Study Group 1997) which compared the costs and outcomes of alternative forms of care. It is imperative that further economic evaluations of primary and secondary prevention and treatment strategies for preterm birth and low birth weight are undertaken if economic information is to be used to inform the efficient allocation of finite resources in this area.

In conclusion, this paper has illustrated the considerable long-term economic impact of preterm birth and low birth weight for various sectors of the economy and for individuals. Many gaps still exist in our understanding of the economic implications of preterm birth and low birth weight. Further, much of the evidence that does exist pertains to specific care delivery patterns that may not be generalisable to different settings. It is imperative that economic considerations are recognized more fully in future studies of preterm birth and low birth weight.

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