Economic implications of multiple births: inpatient hospital costs in the first 5 years of life

J Henderson, C Hockley, S Petrou, M Goldacre, L Davidson


Methods

Oxford record linkage study
This study used data from the ORLS. This is a collection of linked, anonymised records of birth registrations, death certificates, and statistical abstracts of NHS hospital inpatient and day case admissions for a region of southern England. Data collection began in 1966 in Oxfordshire and West Berkshire, and, from 1975, increased its population coverage to include six of the eight districts of the former Oxford Region and, from 1984, the whole of the former Oxford Region. The ORLS had its own data collection systems for maternity and perinatal data until 1989 which covered Oxfordshire and West Berkshire only. Thereafter, data were derived from maternity Hospital Episodes Statistics for the relevant geographical area. Hospital data collection ceased in 1999.

Study population
The study population included all children born to women who both lived and delivered in Oxfordshire or West Berkshire during the period 1 January 1970 to 31 December 1993. Before 1970, much of the relevant perinatal information was missing: a delivery cut off point of 31 December 1993 was required for follow up to cover the first five years of life. Between 1970 and 1993, about 6% of births to residents of Oxfordshire and West Berkshire took place outside of these two areas. These births were not included in the analyses. In addition, hospital admissions occurring outside the ORLS area were not available in the ORLS database and were therefore outside the scope of this study.

Use of hospital services
For each child, a record of inpatient service use between birth and 5 years of age was compiled. Data extracted from the ORLS included the number of babies delivered, date of each...
hospital admission, the duration of hospital stay, specialty on admission, and number of babies in that delivery. Each day case admission was counted as a full 24 hour period for the purposes of this study. A readmission was defined as being any hospital admission after the birth admission. If a baby was admitted directly to neonatal care after birth, this was counted as part of the birth admission. Total time spent in hospital was calculated for each child by summing the lengths of stay of each child's successive admissions. In addition, estimates of days in hospital were calculated for all children who were alive at the start of the period of life of interest (initial birth admission, consecutive years of life, first 5 years), with censoring for deaths. Children who were not readmitted to hospital at all during the first five years of life were included in the denominator in the calculation of means.

**Hospital service costs**

Inpatient costs were calculated for each hospital admission by multiplying the length of stay by the per diem cost of the respective specialty. The specialty based per diem costs were based on the English Department of Health's NHS Trust Financial Returns (TFR2) for 1997–1998 and 1998–1999, which had been averaged over these two financial years to eliminate any random fluctuation in the data. These returns incorporate short run current average revenue costs, plus revenue and capital overheads, and are widely accepted as reliable indicators of hospital service costs. For hospital records with an unknown or incorrect specialty code, the per diem medical or surgical cost was applied, depending on the approximate ORLS code range. All costs are expressed in constant £ sterling (1998–1999 prices) using the NHS Hospital and Community Health Services pay and price deflators provided by the English Department of Health.

**Statistical analysis**

The number of deaths over the first five years of life among live births was examined using Kaplan-Meier analysis. The log rank test was used to compare survival in singleton, twin, and higher order multiple births.

The total duration of hospital admissions, including the initial birth admission, during the first 5 years of life was compared in singletons, twins, and higher order multiple births using a multivariate negative binomial regression. Data on all children were incorporated into this analysis. Relative rates and 95% confidence intervals for the number of days in hospital were calculated after adjustment for duration of life.

Cost differences between singletons, twins, and higher order multiple births that occurred over the first five years, as well as in each of the first five years of life, were tested using multiple linear regression, including only children alive at the start of each period in cost estimates. Costs are reported per child rather than per "set" of twins or triplets. The size of the study sample (276 897) was sufficiently large to expect robust parameter estimates, and therefore, despite the skewed nature of the data, alternative methods such as bootstrapping techniques were not applied.

For all statistical analyses, differences were considered significant if p values were 0.001 or less. This cut off was selected both because multiple comparisons were being made and because in such a large dataset even a small difference tends to be significant. Analyses were performed with a microcomputer using SAS software (version 8.2; SAS Institute Inc, Cary, North Carolina, USA) and SPSS for Windows (release 11.5; SPSS Inc, Chicago, Illinois, USA).

**RESULTS**

The number of babies born as singletons, twins, and higher order multiples in Oxfordshire and West Berkshire between 1970 and 1993 was 270 428, 6284, and 185 respectively. Of the 185 triplet and higher order multiple births, 20 were quadruplets. The rate of multiple births increased slightly over the five years in line with national trends.

Figure 1 illustrates the differential survival of babies born in multiple births. This shows Kaplan-Meier survival up to one year for singletons, twins, and higher order multiple births. Mortality was higher for twins and even greater for higher order multiple births, compared with singleton babies (log rank test \( \chi^2 = 551.4, df = 2, p < 0.0001 \)). Nevertheless, even among higher order multiple births, more than 90% of liveborn infants survived their first year.

As expected, most deaths occurred within the first month of life. Differences in survival decreased over time, and after about six months there were very few deaths.

Babies born in multiple births were at increased risk of both mortality and morbidity. The mean number of days in hospital up to age 5 for singletons, twins, and higher order multiple births respectively were seven, 15, and 30 (table 1). Relative rates, taking account of duration of life, show that twins experienced twice the number of days in hospital as singletons (relative rate 2.40, 95% confidence interval 2.35 to 2.46), and triplets almost eight times as many days in hospital during the first five years of their lives (relative rate 7.58, 95% confidence interval 6.60 to 8.69).

The lengthier periods of inpatient hospital care experienced by babies born in multiple births clearly have financial implications. Table 2 shows estimates based on mean specialty costs. In the birth admission and first year readmissions, the cost of inpatient hospital care increased significantly (\( p < 0.0001 \)) with the multiplicity of birth. Relative to the cost of a singleton baby, the additional cost for

| Table 1 | Days in hospital up to age 5 by multiplicity of birth and relative rates accounting for duration of life |
|-----------------|-------------------------------------------------|-----------------|-----------------|-----------------|-----------------|
|                | Total number | No readmitted | Mean (95% CI) | Median (IQR) | Relative rate | 95% CI |
| Singletons      | 270428       | 126623 (47%)  | 7.1 (7.0 to 7.1) | 5 (4-7) | 1.00            |       |
| Twins           | 6284         | 3137 (50%)   | 14.7 (14.2 to 15.3) | 8 (6-15) | 2.40            | 2.35 to 2.46    |
| Higher order    | 185          | 92 (50%)     | 30.2 (25.6 to 34.8) | 20 (9-39) | 7.58            | 6.60 to 8.69    |
| multiple births |              |              |                |               |                |       |

IQR, Interquartile range; CI, confidence interval.
a twin was £2294 over the first five years. For a baby born in a
higher order multiple birth, the additional cost was £6624.
Annual costs decreased as the children got older, and cost
differences between singletons and multiples tended to
decrease. At ages 2–5 years, there was no significant variation
by multiplicity except during the third year of life when
higher order multiple births had significantly higher inpa-
tient hospital costs. This was largely explained by the
admission to hospital of one set of triplets who required
considerable inpatient support.

**DISCUSSION**

This study shows the increased hospital inpatient costs
associated with twins and higher order multiple births during
the first five years of life. This is in line with work by others,
including Keith et al17 and Callahan and Greene,18 who also
reported increased costs associated with multiple births.
Keith et al17 reported that costs associated with neonatal care
were £3600, £8336, and £60,045 per baby (price date not
reported) for singletons, twins, and triplets respectively.
Callahan and Greene18 reported that the proportion of babies
requiring treatment in neonatal intensive care were 15%, 48%, and 78% for singletons, twins, and triplets respectively,
but that mean lengths of stay in neonatal intensive care were
not significantly different.

The strength of this study is that it is based on a very large,
geographically determined, dataset, which included data on
all inpatient and day case admissions to NHS hospitals
within the former Oxford Region over a 24 year period.
However, it does not include data on outpatient or commu-
nity health service costs or non-health related costs, such as
social service or education related costs. The Mersey study
of low birthweight babies found that costs in all these sectors
were also increased when compared with normal weight babies19, and this is also likely to be the case with low
birthweight multiples. The use of health service resources is
also greater in mothers of multiples both antenatally and
postnatally, although these figures are dwarfed by the costs
of neonatal care.17 None of these data give any measure of the
intangible costs associated with multiple gestation
pregnancies and births.

Although the current study is unusual in covering a five
year time frame, the corollary of this is that the data on births
are not current; the most recent year of hospital data
available from ORLS was 1998, the latest births therefore
were in 1993. Practice may have changed in the intervening
decade.

The ORLS included all admissions to NHS hospitals in the
area covered by the former Oxford Region. Migration out of
this area was estimated at 2.5% per year among the 0–15 age
group.20 This would result in under-reporting of the absolute
levels of resource use and costs. However, there is no
evidence that migration varies by multiplicity of birth, and
therefore the cost differences between groups should be
robust even if the absolute costs are an underestimate.

It is likely that a large proportion of the triplets and higher
order multiple births were conceived with the aid of in vitro
fertilisation, a procedure that has become more common in
the last few decades. Monitoring of assisted reproductive
techniques is carried out by the Human Fertilisation and
Embryology Authority in the United Kingdom. The latter has
recently ruled that not more than two embryos be replaced
during in vitro fertilisation for women aged under 40 years
and not more than three in older women.21 The recommend-
tion to this effect, before the ruling, may have already
reduced the number of multiple births and associated costs to
the NHS, although it is not clear what proportion of multiple
births are due to embryo transfer and what proportion are
due to ovarian stimulation, which may be prescribed by
general practitioners as well as specialists.

This study shows the vast differences in use of hospital
inpatient resources and costs associated with twins, triplets,
and higher order multiple births compared with singletons
during the first five years of life. These costs are only one part
of the picture. A systematic review of the economic
implications of prematurity and low birth weight, including
babies born in multiple births, found that costs of caring for
these children were higher in the community, in social
services, and education; costs to the families were also
greater.17

**What is already known on this topic**

- Babies born in multiple births are more likely to be born
  preterm and at low birth weight.
- Short term health service costs associated with multiple
  births are higher than for singletons.

**What this study adds**

- Mean total number of days in hospital up to age 5
  increases steeply with multiplicity of birth.
- Health service costs up to age 5 likewise increase
  steeply with multiplicity.
- The greatest component of total costs, and the greatest
  difference in costs, occurs in the first year of life.

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**Table 2 Mean cost of hospital inpatient stay by multiplicity**

<table>
<thead>
<tr>
<th>Age at admission</th>
<th>Singletons</th>
<th>Twins</th>
<th>Higher order multiple births</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean costs</td>
<td>95% CI</td>
<td>No of children</td>
</tr>
<tr>
<td>Initial birth admission</td>
<td>846</td>
<td>841 to 850</td>
<td>270428</td>
</tr>
<tr>
<td>Readmissions starting in:</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1st year</td>
<td>414</td>
<td>403 to 427</td>
<td>269596</td>
</tr>
<tr>
<td>2nd year</td>
<td>104</td>
<td>101 to 107</td>
<td>268318</td>
</tr>
<tr>
<td>3rd year</td>
<td>74</td>
<td>71 to 77</td>
<td>268184</td>
</tr>
<tr>
<td>4th year</td>
<td>67</td>
<td>64 to 70</td>
<td>268099</td>
</tr>
<tr>
<td>5th year</td>
<td>65</td>
<td>62 to 68</td>
<td>268034</td>
</tr>
<tr>
<td>Entire study period</td>
<td>1532</td>
<td>1516 to 1548</td>
<td>270428</td>
</tr>
</tbody>
</table>

Mean costs are per child in £ sterling.
*Only children alive at the start of the period were included in the cost estimates.
†Mean costs were adjusted for duration of life as a categorical variable grouped in 0–7 days, 8–28 days, 29–365 days, and 366–1825 days.
There is scope for more in-depth research into the costs borne by families, including intangible costs, associated with multiple births. Policy implications of increasing provision of assisted reproductive techniques, and in vitro fertilisation in particular, need to be assessed. Draft guidelines from the National Institute for Clinical Excellence (NICE) propose recommending that, for couples meeting certain criteria, up to three cycles of in vitro fertilisation be paid for by the NHS. This would address some of the problems of the “postcode lottery” but would work against local decision making. If the draft guidelines are agreed, it would be important to evaluate outcomes from clinical, psychological, and health economic perspectives.

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