Congenital anterior abdominal wall defects in England and Wales 1987-93: retrospective analysis of OPCS data

Kelvin H Tan, Mark D Kilby, Martin J Whittle, Bryan R Beattie, Ian W Booth, Beverley J Botting

See editorial by Chitty

Abstract

***Objectives*—Analysis of incidence and characteristics of congenital abdominal wall defects, with special reference to the differences between the incidence of gastroschisis and exomphalos (omphalocele).**

***Design*—Retrospective analysis using data from the Office of Population Censuses and Surveys (recoded to differentiate exomphalos and gastroschisis) and the National Congenital Malformation Notification Scheme.**

***Setting*—England and Wales, 1987 to 1993.**

***Results*—1043 congenital anterior abdominal wall defects were notified within the seven year study period. Of these, 539 were classified as gastroschisis, 448 as exomphalos, 19 as "prune belly syndrome," and 37 as "unclassified." Gastroschisis doubled in incidence from 0.65 in 1987 to 1.35 per 10 000 total births in 1991, with little further change; the incidence of exomphalos decreased from 1.13 to 0.77 per 10 000 births. The overall incidence of notified congenital abdominal wall defects was 2.15 per 10 000 total births. Gastroschisis was associated with a lower overall maternal age than exomphalos and with a significantly lower proportion of additional reported congenital malformations (5.0%) than in the cohort with exomphalos (27.4%) (odds ratio 0.14, 95% confidence interval 0.09 to 0.22; P<0.001). The sex ratio of the two cohorts was the same. The incidence of gastroschisis and exomphalos was higher in the northern regions of England than in the south east of the country.**

***Conclusions*—The national congenital malformation notification system showed an increasing trend in the incidence of fetuses born with gastroschisis and a progressive decreasing incidence of exomphalos in England and Wales between 1987 and 1993. Although the reasons for this are likely to be multifactorial, a true differential change seems likely. The observed increase in incidence of gastroschisis relative to exomphalos and the differentiation in maternal age have implications for resource management within the NHS and warrant further epidemiological monitoring. Regional differences may be due to a dietary or environmental factor, which requires further study.**

Division of Fetal Medicine, Academic Department of Obstetrics, University of Birmingham, Birmingham B15 2TG
Kelvin H Tan, *research registrar*
Mark D Kilby, *senior lecturer*
Martin J Whittle, *professor*

Department of Fetal Medicine, University of Wales Heath Park, Cardiff CF 4 4XN
Bryan R Beattie, *consultant*

Institute of Child Health, University of Birmingham
Ian W Booth, *professor*

Office for National Statistics, London WC2B 6JP
Beverley J Botting, *medical statistician*

Correspondence to:
Dr Kilby.

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Introduction

Uncertainty and confusion exist concerning the embryology, clinical presentation, and management of congenital anterior abdominal wall defects.[1-5] Gastroschisis (fig 1) has been considered a rare malformation, in contrast with exomphalos (fig 2),[6] although the incidence of gastroschisis has risen in the past two decades without a corresponding change in the number of neonates with exomphalos.[7-13] We therefore investigated the epidemiology of congenital abdominal wall defects in England and Wales between 1987 and 1993, with special reference to the characteristics, incidences, and regional differences between gastroschisis and exomphalos. We also present data on the incidence of terminations of pregnancy in England and Wales for anterior abdominal wall defects during this time.

Fig 1—*Gastroschisis, one of the major congenital anterior abdominal wall defects, is a paraumbilical herniation of gastrointestinal structures into the amniotic cavity*

Patients and methods

This retrospective, observational study used data collected by the Office of Population Censuses and Surveys (OPCS; known since its merger with the Central Statistics Office on 1 April 1996 as the Office for National Statistics) on notifications of congenital abdominal wall defects, congenital structural malformations, and birth registrations for the years between 1987 and 1993. In the OPCS congenital malformation notification scheme, a standard record form (SD56) is completed by district health authorities to notify details of each malformed live birth or stillbirth. Only malformations detected at delivery or within 10 days of birth were included during 1987-93. The form requested a textual description of all the observed malformations, and up to eight different malformations for each fetus were coded by trained OPCS staff according to the 9th revision of the *International Classification of Diseases* (ICD-9). Unfortunately, as the original coding was "anterior abdominal wall defects" these data did not differentiate between gastroschisis and exomphalos. For reanalysis, the original form was extracted (with year of report blinded) and a new code given to indicate gastroschisis or exomphalos.

A total of 1043 congenital anterior abdominal wall defects were notified to OPCS out of an estimated 4 859 221 total births (abortion data not included) between 1987 and 1993 in England and Wales. Additional information on births, legal terminations, and notified congenital abnormalities was obtained from OPCS publications.[15 16]

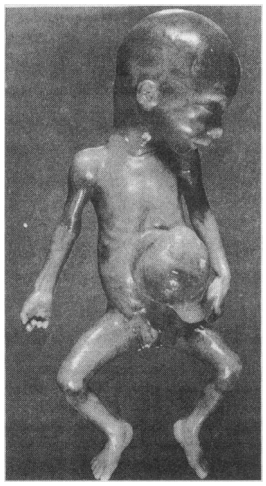

Fig 2—*In exomphalos, gastrointestinal structures protrude into the cord root. Reproduced by permission of Blackwell Scientific Publications*[14]

STATISTICAL ANALYSES

Intergroup comparisons for continuous variables with a non-parametric distribution were made using the Mann-Whitney U test to determine significant differences between the data sets. For such data, median values and interquartile ranges are described. Categorical data were analysed using Fisher's exact test and odds ratios and 95% confidence intervals. Significance was taken as P<0.05 unless otherwise stated.

Results

Of 1043 congenital anterior abdominal wall defects notified to the OPCS register, 539 (51.7%) were classified as gastroschisis, 448 (42.9%) exomphalos, and 19 (1.8%) "prune belly" syndrome; 37 (3.5%) were "unclassified." During 1987-93 an estimated 4 859 221 births occurred in England and Wales, giving an incidence for anterior abdominal wall defects of 2.15 per 10 000 births. When this was stratified into gastroschisis and exomphalos the mean incidences were 1.11 and 0.92 respectively. Table 1 shows the total number of terminations performed between 1987 and 1993 for each of the ICD-9 codes for anterior abdominal wall defects (550-553; 756-7). The annual total remained remarkably constant during this time. We could not differentiate the two types of defect, but exomphalos would be more likely to end in termination of pregnancy because of coexistent anomalies.

Figure 3 shows changes in incidence and trends in the data for individual years between 1987 and 1993.

Table 1—*Terminations of pregnancy in England and Wales 1987-93 for congenital anterior abdominal wall defects*

Year	No of terminations: Codes 550-553*	Code 757*	Total
1987	2	27	29
1988	5	22	27
1989	10	30	40
1990	4	32	36
1991	9	26	35
1992	15	33	48
1993	8	36	44

*ICD-9 codes: 550 Anterior abdominal wall hernia (inguinal); 551 Other hernia of the abdominal cavity and gangrene; 552 Other hernia of the abdominal cavity and obstruction; 553 Other hernia of the abdominal cavity with no evidence of gangrene/obstruction; 757 Congenital abdominal cavity integument

Table 2—*Characteristics of notified cases of gastroschisis and exomphalos in England and Wales, 1987-93. Data not presented as proportions are shown as median (interquartile range)*

	Gastroschisis (n = 539)	Exomphalos (n = 448)	Odds ratio (95% confidence interval)	P value
Incidence (/10 000 births)	1.11	0.92	1.20 (1.06 to 1.36)	<0.01
Maternal age (years)	21.0 (19-25)	26.0 (22-30)		<0.001
% Of mothers aged under 20	34.1	13.0	3.45 (2.48 to 4.79)	<0.001
% Nulliparous mothers	70.1	52.5	2.11 (1.61 to 2.78)	<0.001
Birth weight (g)	2332 (2008-2750)	2625 (2070-3280)		<0.001
Gestation at birth (weeks)	37.0 (35-38)	37.0 (35-39)		>0.05
% Small for dates (<5th centile for gestational age)	31.9	23.7	1.51 (1.10 to 2.08)	<0.01
% Boys	52.7	52.7	1.00 (0.78 to 1.28)	>0.5
% Twins	1.7	3.8	0.43 (0.19 to 0.98)	<0.05
% With associated anomalies*	5.0	27.4	6.42 (4.21 to 9.80)	<0.001
% Stillborn	3.5	11.0	0.30 (0.17 to 0.53)	<0.001
% Of early neonatal deaths	3.1	10.8	0.28 (0.15 to 0.53)	<0.001
Perinatal mortality (/1000 births)	65	206	0.32 (0.21 to 0.48)	<0.001

*Some babies were born with multiple abnormalities (but counted as single affected fetus).

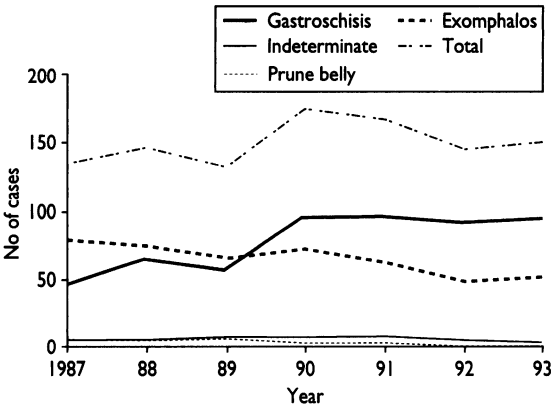

Fig 3—*Number of notified cases of congenital abdominal wall defects in England and Wales, 1987-93*

Gastroschisis showed a rising trend, from 46 notified cases in 1987 to 95 cases in 1991, with little further increase in 1992-3. Thus, the incidence of gastroschisis rose rapidly from 0.65 (in 1987) to 1.35 per 10 000 total births in 1991. Thereafter, the incidence seemed to change little from 1.39 per 10 000 births in 1993 (χ^2 for trend = 34.5, df = 6; P<0.0001).

In contrast, the total numbers of births with exomphalos fell from 78 cases in 1987 to 52 in 1993, so that the incidence fell from 1.13 to 0.77 per 10 000 total births between 1987 until 1993 (χ^2 for trend = 8.75, df = 1; P<0.005). The notified cases of "prune belly" syndrome decreased from 6 cases in 1987 to zero in 1993 (probably due to reclassification under early urethral obstruction sequence); while the indeterminate/unspecified cases of congenital abdominal wall defect remained relatively constant at between 3 to 7 cases a year.

Table 2 shows the characteristics of the cohorts of gastroschisis and exomphalos. The incidence of gastroschisis was significantly associated with decreasing maternal age (table 3). Congenital structural abnormalities were significantly more likely to be associated with exomphalos (27.4%) than gastroschisis (5%) (odds ratio 6.42 (95% confidence interval 4.21 to 9.80); P<0.001). The range of anomalies associated with exomphalos varied more widely than in the gastroschisis cohort.

The median birth weight of babies with gastroschisis was significantly lower than those with exomphalos (2332 (2008 to 2750) g *v* 2625 (2070 to 3280) g; P<0.001). However, babies born with gastroschisis had a narrower interquartile range than those with exomphalos (742 g *v* 1210 g), which was reflected in the interquartile ranges of the gestations of both groups (3 weeks *v* 4 weeks). Matched for gestational age, babies with gastroschisis were significantly smaller than babies with exomphalos (31.9% *v* 23.7% were below the 5th centile for birth weight; odds ratio 1.51 (1.10 to 2.08); P<0.01). The perinatal mortality rate of babies with gastroschisis (65/1000) was significantly lower than those pregnancies complicated by exomphalos (206/1000; odds ratio 0.32 (0.21 to 0.48); P<0.001).

The incidence of congenital abdominal wall defects was higher in the regions of northern England than in regions in south east England. Stratification of data into gastroschisis and exomphalos showed this directional regional variations in both conditions. The incidence of total abdominal wall defects, gastroschisis, and exomphalos in the northern regions was twice that in South East Thames Region during the study period (table 4; P<0.01). However, the total number of legal abortions reported for known or suspected fetal anomaly was higher in the Northern region (32.9/10 000) than in the south (19.6/10 000 in South East Thames), so the regional differences are unlikely to be secondary to the number of abortions performed.

Table 3—*Incidence of gastroschisis and exomphalos per 10 000 total births related to mother's age, England and Wales, 1987-93*

Age range	Mean total births annually*	Gastroschisis		Exomphalos	
		No	Incidence	No	Incidence
<20	54 905	181	4.71	58	1.51
20-24	182 342	216	1.69	115	0.90
25-29	246 237	99	0.57	140	0.81
30-34	151 887	28	0.26	84	0.79
35-39	51 304	5	0.14	35	0.97
>40	9 546	2	0.30	13	1.95
Total	699 220	531	1.09	445	0.91
Not stated		8		3	

*Based on total births 1987-92.

Examination of the numbers of abortions performed for anterior abdominal wall defects annually between 1987 and 1993 showed that they were remarkably constant over this period, with only a slight increase towards the end (table 1). Series DH3 publications give figures on the number of total births by area of residence and maternal age[17], and these figures show an increase from south to north in the United Kingdom in the number of women under 20 giving birth (table 5). This finding may be partly responsible for the increased incidence of gastroschisis in the north of England, although both gastroschisis and exomphalos show the same regional, but not age specific, trend.[18-21]

Discussion

The results of this observational study are consistent with others indicating the rise in the incidence of gastroschisis worldwide and with data from England which indicate that the incidence of gastroschisis has increased threefold over 25 years.[22] Our data show a remarkably rapid rise in incidence of gastroschisis, from 0.65 to 1.11 per 10 000 births over the seven years of the study; in contrast with other series,[10 13 23 24] they show that the incidence of gastroschisis exceeded that of exomphalos (fig 3).

It seems unlikely that this apparent rise in the incidence of gastroschisis would be explained by a greater tendency to termination of pregnancies with gastroschisis in the early part of this study, and if anything there was a slight increase in termination for abdominal wall defects towards the end of the study period. However, the falling incidence of exomphalos may well be the result of termination because of associated prenatally diagnosed anomalies. However, in January 1990 the Office of Censuses and Surveys introduced an exclusion list, which included paraumbilical hernia. The overall number of notifications fell between 1989 and 1990, and this may be accounted for by the introduction of this list, which may have had an effect on abdominal wall defects. This would be unlikely to have influenced the reported cases of gastroschisis during this period (or if it did it would be likely to reduce overall notifications rather than to increase them).

Our series confirms the association between gastroschisis and low maternal age.[9 10 25-28] Younger mothers are significantly more likely than women older than this age to have a child with gastroschisis (table 3). Interestingly, this age relation does not seem to exist for women whose babies have exomphalos.

Table 4—*Incidence (rates per 10 000 total births[20]) of live births, stillbirths, and perinatal deaths in mothers under 20 by area of residence, 1991*

Regional health authority	Stillbirths	Perinatal deaths	Live births
Northern	41	112	990
Yorkshire	57	109	920
Trent	63	103	870
North East Thames	50	94	600
South East Thames	78	134	620

Table 5—*Incidence of abdominal wall defects per 10 000 births in regions of England and Wales, 1987-93. Estimate of denominator is based on total births 1987-93 in region*

Region	Incidence per 10 000 births		
	All abdominal wall defects	Gastroschisis	Exomphalos
Northern	3.11	1.55	1.38
Yorkshire	2.24	1.11	0.96
North West	2.86	1.60	1.13
Mersey	2.09	1.32	0.64
Trent	3.09	1.62	1.23
West Midlands	1.84	0.87	0.89
Oxford	1.59	0.76	0.76
East Anglia	2.00	0.98	0.98
North West Thames	1.80	0.65	1.01
North East Thames	1.72	0.87	0.74
South East Thames	1.47	0.72	0.64
South West Thames	1.23	0.58	0.62
Wessex	2.56	1.58	0.90
South West	2.28	1.40	0.77
Wales	1.97	0.96	0.96

REGIONAL DIFFERENCES

Both gastroschisis and exomphalos have a higher incidence in regions towards the north of England, a trend which is as distinct as those shown for neural tube defects over a similar period (1987-91) in England and Wales.[16] There are regional differences in the number of live births to women under the age of 20 (table 4).[17] Although such a demographic distribution may in part explain regional differences in the incidence of gastroschisis, it is unlikely to be the whole explanation. Indeed, the incidence of gastroschisis is some 111% greater in the northern than southern regions, whereas the corresponding increase in live births to women under 20 was only 59%. These regional data are consistent with studies from the west of Scotland which indicated that anterior abdominal wall defects had an incidence of 4 per 10 000 total births, much higher than that of England and Wales.[29]

A factor which may influence regional incidences of congenital malformations, including anterior abdominal wall defects, is that of the differing regional uptake of abortion services, as total legal abortions amounted to 24.1% of approximately 3.5 million births between 1987 and 1991.[15] The total number of legal abortions within this five year period showed an increased incidence in the south east of England (Northern Region 1712/10 000 births; South East Thames 3114/10 000 births). This may have the effect of reducing the incidence of gastroschisis, as abortions in teenagers are increasing.[30] However, examination of the data for the incidence of abortion for known or suspected fetal abnormality indicates a trend similar to that for the incidence of anterior abdominal wall defects in England and Wales. It seems improbable that the total number of pregnancies with exomphalos terminated by chance would be greater than those with gastroschisis. It is thus unlikely that the change in regional incidences of congenital anterior abdominal wall defects is directly attributable to changes in abortion rates.

CAUSES

It is extremely doubtful that a genetic cause is responsible for gastroschisis, as this abnormality seems to occur sporadically and have a low recurrence rate, although the possibility of a nutritional or enviromental aetiology remains.[31-33] Over the past five years, the inci-

Key messages

- Gastroschisis doubled in incidence from 0.65 per 10 000 total births in 1987 to 1.35 per 10 000 in 1991, with little further change between 1992 and 1993; the incidence of exomphalos decreased from 1.13 to 0.77 per 10 000 births over the same period
- Gastroschisis was also associated with a significantly lower proportion of additional reported congenital malformations
- Younger mothers are significantly more likely than older mothers to have a child with gastroschisis
- The incidence of congenital abdominal wall defects seemed to be higher in the regions in the north than in the south east of England

dence of cigarette smoking has increased in pregnant teenagers compared with women 20 and over.[34] The clustering of gastroschisis in younger mothers and the observed increase in incidence of this malformation over the past seven years indicates a possiblity of an association between cigarette consumption and an interruption of fetal omphalomesenteric arterial blood supply.[35] Few estimates of drug taking in adolescent pregnant women have been undertaken. However, an increased risk for gastroschisis has been described in women using recreational drugs before or in early pregnancy, with cocaine being associated with a fourfold increase in the risk; using marijuana, amphetamines, or alcohol doubles the risk.[36-38]

NOTIFICATION

The OPCS system of voluntary notification has always had several deficiencies which have been largely identified and addressed.[39] These include the under-notification and misclassification of major congenital malformations, underascertainment of additional malformations, and overnotification of trivial malformations.[18 19] Congenital anterior abdominal wall defects should be readily apparent and diagnosed at birth and therefore should be reliably ascertained by the OPCS system, in contrast with conditions which are "internal" malformations.[19-21] Data from the west Midlands, however, indicate that 65 babies with gastroschisis were operated on by the paediatric surgical unit between 1987 and 1993, whereas 46 cases were reported to OPCS, indicating undernotification (V Upadhyay *et al*, British Paediatric Association, York, 1995).

National data from the OPCS (now the Office for National Statistics) could further be enhanced by improving the census quality (in particular ensuring the compulsory notification of fetal abnormalities, especially in conjunction with regional perinatal and malformation registries) and the inclusion of all data from abortions carried out following prenatal diagnostic procedures. The implementation of such change is vital for improved epidemiological and management strategies that may have considerable implications for the provision of resources in obstetric and paediatric specialties.

When the deficiencies of the current national notification system for congenital abnormalities are taken into account, these national data from 1987 to 1993 show an increasing incidence of fetuses born with gastroschisis while data for exomphalos showed a slowly decreasing trend. These data are consistent with our data from the West Midlands region (V Upadhyay *et al*, 1995) and from the California birth defects monitoring programme.[37 38] Although this could be due to better differentiation between the two groups, selective reporting of anterior abdominal wall defects, and increased termination of exomphalos, a true differential change seems likely. The increased incidence of gastroschisis not only poses important implications for obstetric and paediatric surgical management but demands more refined epidemiological monitoring (through further improvement in the national and regional notification systems) and requires further research into causation.

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