Journal of Pediatric Surgery (2010) **45**, 1808–1816

ELSEVIER

Journal of Pediatric Surgery

www.elsevier.com/locate/jpedsurg

Gastroschisis: a national cohort study to describe contemporary surgical strategies and outcomes☆,☆☆,★

Anthony Owen[a], Sean Marven[a], Paul Johnson[b], Jennifer Kurinczuk[c], Patsy Spark[c], Elizabeth S. Draper[d], Peter Brocklehurst[c], Marian Knight[c,*] on behalf of BAPS-CASS

[a]*Paediatric Surgical Unit, Sheffield Children's Hospital NHS Foundation Trust, S10 2TH Sheffield, UK*
[b]*Department of Paediatric Surgery, University of Oxford, OX3 9DU Oxford, UK*
[c]*National Perinatal Epidemiology Unit, University of Oxford, OX3 7LF Oxford, UK*
[d]*Department of Health Sciences, University of Leicester, LE1 6TP Leicester, UK*

Received 4 November 2009; revised 20 January 2010; accepted 28 January 2010

Key words:
Gastroschisis;
Abdominal wall defects;
Surveillance;
Outcomes;
Population-based

Abstract

Background: Information on adoption of newer surgical strategies for gastroschisis and their outcomes is largely limited to hospital-based studies. The aim of this study was to use a new UK national surveillance system to identify cases and thus to describe the contemporary surgical management and outcomes of gastroschisis.

Methods: We conducted a national cohort study using the British Association of Paediatric Surgeons Congenital Anomalies Surveillance System to identify cases between October 2006 and March 2008.

Results: All 28 surgical units in the United Kingdom and Ireland participated (100%). Data were received for 95% of notified cases of gastroschisis (n = 393). Three hundred thirty-six infants (85.5%) had simple gastroschisis; 45 infants (11.5%) had complex gastroschisis. For 12 infants (3.0%), the type of gastroschisis could not be categorized. Operative primary closure (n = 170, or 51%) and staged closure after a preformed silo (n = 120, or 36%) were the most commonly used intended techniques for simple gastroschisis. Outcomes for infants with complex gastroschisis were significantly poorer than for simple cases, although all deaths occurred in the simple group.

Conclusions: This study provides a comprehensive picture of current UK practice in the surgical management of gastroschisis. Further follow-up data will help to elucidate additional prognostic factors and guide future research.

☆ Contributions: A.O. assisted with the design of the study, carried out some analysis, and wrote a first draft of the paper. S.M. had the original idea for the investigation of management of gastroschisis, assisted with the design of the study, provided clinical input, and contributed to the analysis and writing the paper. P.J. provided clinical input to the study and contributed to the analysis and writing of the paper. J.J.K., E.D., and P.B. assisted with the design of the study and contributed to the analysis and writing of the paper. P.S. assisted with data coding, conducted validation of the data and the analysis, and contributed to the writing of the paper. M.K. designed the study, coordinated data collection, coded the data, and supervised the analysis, writing, and editing of the paper.

☆☆ Funding: Funding for the study was provided by the charity BDF Newlife and the British Association of Paediatric Surgeons.

★ Conflicts of interest: None to declare.

* Corresponding author. Tel.: +44 1865 289727; fax: +44 1865 289701.
E-mail address: marian.knight@npeu.ox.ac.uk (M. Knight).

0022-3468/$ – see front matter © 2010 Elsevier Inc. All rights reserved.
doi:10.1016/j.jpedsurg.2010.01.036

The contemporary surgical strategies for the correction of gastroschisis have evolved considerably in modern times. The principles of management however remain the same, first, to reduce the viscera safely and, second, to close the abdominal wall defect with an acceptable cosmetic appearance. In addition, nutrition must be supported and associated anomalies or complications identified and managed appropriately.

The strategies that have been developed are often summarized as primary or staged procedures, either of which may or may not be performed under general anesthesia, that is, operatively or nonoperatively. Operative primary reduction with sutured fascial defect closure has become the standard initial surgical strategy, whereas operative staged reduction is frequently used as a rescue strategy when reduction is deemed unsafe or physically impossible because of visceroabdominal disproportion. Operative staged reduction has been predominantly achieved by suturing a synthetic material to the enlarged defect and delayed defect closure. This amounts to an individualized or selective approach and has been the acceptable standard for more than 20 years, after an era where operative staged reduction was considered to be the safest approach but with poorer cosmetic outcome. In sharp contrast, 2 nonoperative strategies have emerged; "ward reduction" described by Bianchi and Dickson [1] in 1998 and more recently preformed silos for routine staged reduction also without general anesthesia which have challenged the standard approaches [2,3]. After reduction of the viscera, subsequent defect closure is achieved in 1 of 3 main ways: operative suture closure approximating the fascia usually under general anesthesia, nonoperative dressing closure that relies on defect contraction, or prosthetic patch closure, which may be temporary or permanent using either synthetic or biologic materials [4,5].

Information on the contemporary usage of these strategies or adoption of the newer techniques and their outcomes is limited to hospital or single-operator–based studies [2,6]. There is no national information describing the frequency with which these different techniques are used in the United Kingdom nor data concerning the characteristics of affected infants or their outcomes.

In 2005, the Chief Medical Officer in the United Kingdom issued a report, "Gastroschisis: A Growing Concern" [7], which recommended research into the rising prevalence and the management of gastroschisis. In response, a collaboration between the British Association of Paediatric Surgeons (BAPS) and the National Perinatal Epidemiological Unit was forged to establish the British Association of Paediatric Surgeons Congenital Anomalies Surveillance System (BAPS-CASS), a national system, to conduct population-based studies of a range of conditions requiring pediatric surgical management in the United Kingdom. The aim of this study was to describe the contemporary surgical management of gastroschisis throughout the United Kingdom and associated perioperative outcomes.

1. Methods

1.1. Case definition

We defined cases as any liveborn infant with gastroschisis, defined as a congenital malformation characterized by visceral herniation through an abdominal wall defect lateral to an intact umbilical cord and not covered by a membrane. We excluded infants with aplasia or hypoplasia of abdominal muscles, skin-covered umbilical hernia, and exomphalos.

1.2. Data collection

We identified cases through BAPS-CASS between October 2006 and March 2008 inclusively. We identified a nominated reporting clinician in each of the 28 pediatric surgical centers in the United Kingdom and Ireland. Each month, we sent them a routine monthly reporting card, requesting details of the number of infants born and admitted to their unit that month with gastroschisis. We also asked participating clinicians to return a "nil report," that is, a card noting that there were no cases, in order that we could be confident that all cases were being reported. If a clinician did not return a card for 3 successive months, we contacted them by telephone or e-mail to obtain the missing case reports.

In response to a monthly card returned indicating that there had been a case of gastroschisis, we sent clinicians a form requesting further details of the infant, including diagnosis, surgical management, operative complications, and other outcomes. If the form was not returned after 6 weeks, we sent a reminder letter; after 10 weeks, we contacted the clinician by e-mail; after 14 and 28 weeks, we sent a further reminder including a new form. This form sought details of outcomes up to first discharge from hospital or 6 months of age, whichever came sooner.

We double entered all data into a customized database and excluded duplicate reports by comparing hospital, mother's year of birth, and date of first operation. If any data items were missing or fell outside prespecified ranges, we contacted the clinician who had completed the form to complete the information. If we did not receive a response to our first request for missing information, we sent the request again a month later and then again after 2 and 3 months.

Note that details of 26 patients from Sheffield Children's Hospital and 13 from King's College Hospital have been reported previously as part of a separate retrospective study of all cases of gastroschisis managed with a preformed silo in 4 UK neonatal surgical units (Sheffield Children's Hospital, Southampton University Hospitals Trust, King's College Hospital, London, and John Radcliffe Hospital, Oxford) between January 2001 and December 2007 [8].

1.3. Additional case ascertainment

In addition to identifying cases through BAPS-CASS, we also identified cases through the UK Obstetric Surveillance System and the British Isles Network of Congenital Anomalies Registers.[8,9] Where we identified a case that had not been reported through BAPS-CASS, we contacted the relevant BAPS-CASS reporting clinician and asked them to complete a data collection form.

1.4. Ethics committee approval

This study was approved by the London Multi-centre Research Ethics Committee (Study reference 05/MRE02/82).

1.5. Statistical analyses

We compared outcomes in infants with simple gastroschisis (defined as intact continuous bowel that is not compromised or breached at delivery or presentation) and those with complex gastroschisis (defined as the presence of 1 or more of the following criteria: intestinal atresia, perforation or intestinal necrosis at delivery or presentation, or missed atresia) using nonparametric methods. We calculated risk ratios (RRs) with 95% confidence intervals (CIs). All analyses were carried out using SPSS version 15 (SPSS, Chicago, IL) and STATA version 10 (StataCorp, College Station, TX).

2. Results

All 28 pediatric surgical units in the United Kingdom and Ireland returned data to BAPS-CASS: 100% participation. Data were received for 95% of notified cases of gastroschisis (Fig. 1). We received data for a total of 393 cases of gastroschisis in an estimated birth cohort of 1.1 million infants during the 18 months of the study.

2.1. Infant characteristics

Three hundred thirty-six infants (85.5%) had simple gastroschisis; 45 infants (11.5%) had complex gastroschisis, and for 12 infants (3.0%), the type of gastroschisis could not be categorized (Table 1). There were no cases of closed gastroschisis. Of the 45 cases of complex gastroschisis reported, 31 infants had atresia only, 12 infants had atresia and bowel perforation, and 2 patients had bowel perforation only. Infants with complex gastroschisis were significantly more likely to be born preterm or with a birth weight less than 2500 g ($P < 0.05$). Other characteristics of infants with both simple and complex gastroschisis are shown in Table 1.

Ninety-eight percent of all cases of gastroschisis (n = 385) were detected antenatally; 97% had the defect sited to the right of the umbilicus. The size of the defect was measured in 28% of infants, with a median reported diameter of 3 cm (range, 0.3-7 cm). The bowel length was only measured in 3% of cases.

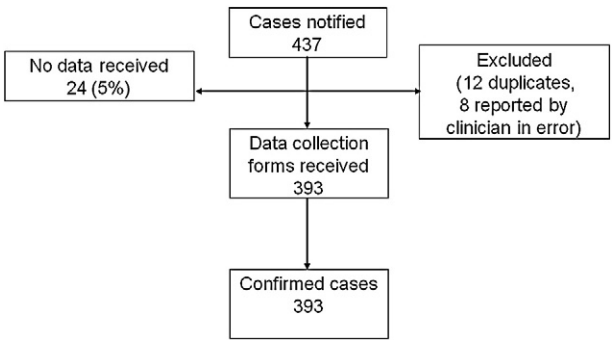

Fig. 1 Case reporting and completeness of data collection.

Table 1 Patient characteristics according to type of gastroschisis

Characteristic	Simple, n (%)[a] (n = 336)	Complex, n (%)[a] (n = 45)	Unknown, n (%)[a] (n = 12)
Ethnicity			
White	297 (91)	38 (90)	8 (89)
Nonwhite	28 (9)	4 (10)	1 (11)
Sex			
Male	157 (47)	16 (36)	4 (36)
Female	179 (53)	29 (64)	7 (64)
Birth weight (g)			
≥2500	145 (43)	12 (27)	4 (44)
<2500	191 (57)	33 (73)	5 (56)
Gestational age (wk)			
≥37	192 (57)	12 (27)	4 (44)
<37	144 (43)	33 (73)	5 (56)
Antenatally diagnosed			
No	4 (1)	1 (2)	0 (0)
Yes	332 (99)	44 (98)	9 (100)
Associated anomalies (nonbowel)			
No	316 (94)	43 (96)	7 (88)
Yes	20 (6)	2 (4)	1 (12)
Mode of delivery			
Caesarean section before labor	41 (12)	11 (24)	1 (9)
Cesarean section after labor onset	86 (26)	10 (22)	2 (18)
Other	209 (62)	24 (53)	8 (73)
Transferred after birth			
No	221 (66)	29 (64)	5 (45)
Yes	115 (34)	16 (36)	6 (55)

[a] Percentages of those with data.

2.2. Surgical management

The operative management (primary reduction and defect closure and staged reduction with delayed defect closure) of

the simple gastroschisis cases is shown in Fig. 2 and Table 2. Operative primary reduction and defect closure (n = 170, or 51%) and staged reduction with delayed defect closure using an application of a preformed silo (n = 120, or 36%) were the most commonly used intended techniques.

For infants with simple gastroschisis where the intended initial management with a preformed silo was successful (n = 113), silo removal was performed after a median of 5 days (range, 0-26 days).

The operative management (primary and staged closure) of the complex gastroschisis cases is shown in Fig. 3 and Table 3. Operative primary reduction and defect closure (n = 32, or 71%) and staged reduction with delayed defect closure by application of a preformed silo (n = 11, or 24%) were the most common techniques.

Table 2 Initial management for simple gastroschisis

Initial management, n (%) (n = 336)	Successful, n (%)	Converted, n (%)
Operative primary closure, 170 (51)	140 (82)	30 (18)
Ward reduction, 25 (7)	18 (72)	7 (28)
Preformed silo, 120 (36)	113 (94)	7 (6)
Custom silo, 15 (4)	14 (93)	1 (7)
Prosthetic patch, 5 (1)	5 (100)	0 (0)
Other primary closure, 1 (0.3)	1 (100)	0 (0)

2.3. Outcomes

The outcomes for both simple and complex gastroschisis are shown in Table 4. Infants with complex gastroschisis were more likely to be ventilated postoperatively (RR, 1.21; 95% CI, 1.09-1.33), more likely to require reoperation (RR, 6.53; 95% CI, 4.70-9.09), more likely to develop intestinal failure associated liver disease (IFALD; RR, 8.21; 95% CI, 3.70-18.2), and more likely to receive Total Parenteral Nutrition (TPN) for more than 28 days (RR, 2.07; 95% CI, 1.71-2.51). There were no other statistically significant differences in any of the outcomes we examined.

Outcomes for the 2 most frequently performed intended procedures for simple gastroschisis, operative fascial closure, and preformed silo are shown in Table 5. Thirty-four of the infants with simple gastroschisis managed with either of these techniques had reoperations; 25 had 1 reoperation, and 12 had more than 1 further operations.

Six infants died in the neonatal period, all of whom had simple gastroschisis, 3 from bowel ischemia and associated complications of gastroschisis after initial surgical management with either preformed (n = 2) or custom silo (n = 1), 1 caused by respiratory complications of prematurity, 1 caused by multiple congenital anomalies, and 1 from unrelated infection.

3. Discussion

This study describes, for the first time, the contemporary surgical management and associated outcomes of a UK national cohort of infants with gastroschisis. This has been achieved through a unique collaboration of all tertiary pediatric surgical centers in the United Kingdom and Ireland, the BAPS-CASS, which enables national cohort studies of a range of pediatric surgical conditions. Typically, studies of such conditions report the surgical outcomes of hospital-based case series; hospital-based case series are subject to biases that limit their validity and generalizability, including particularly case selection and information bias and the biases inherent in single-operator series. By collecting information about all cases nationally, the information generated from this study is uniquely generalizable, providing a true picture of current surgical management of

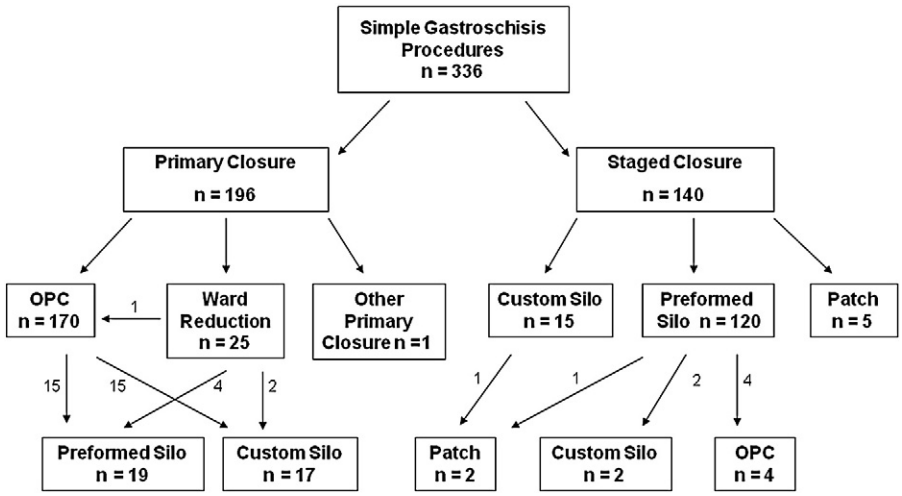

Fig. 2 Initial management for simple gastroschisis. Numbers indicate converted cases and subsequent management. OPC indicates operative primary reduction with sutured fascial defect closure.

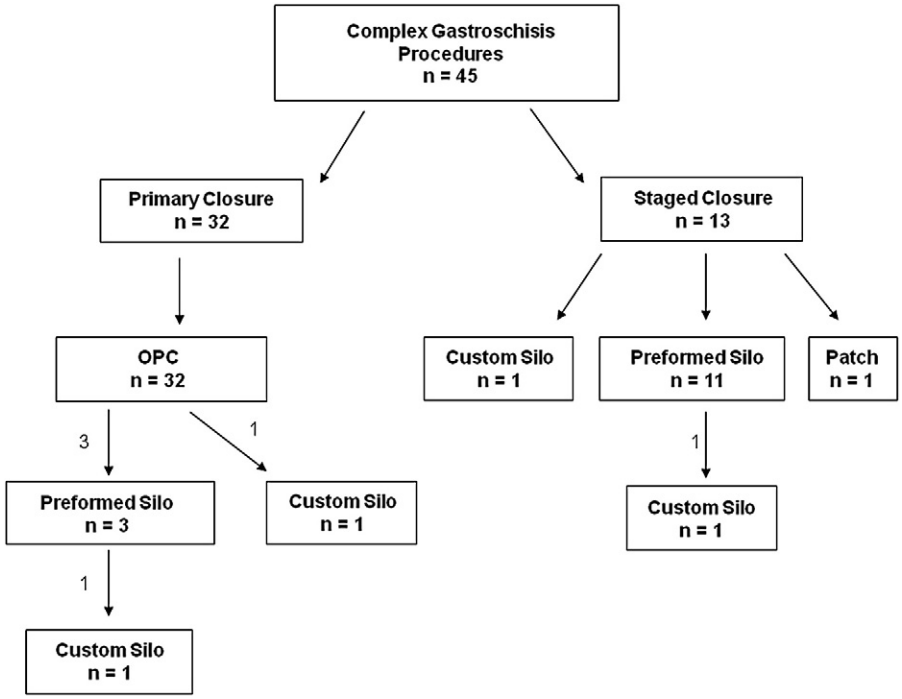

Fig. 3 Initial management for complex gastroschisis. Numbers indicate converted cases and subsequent management. OPC indicates operative primary reduction with sutured fascial defect closure.

gastroschisis on a population basis and thus a benchmark against which the practice and outcomes in single centers may be compared, as well as a baseline against which any future changes in practice or service provision may be evaluated. Center-based studies without a clearly defined population base are not able to be used for these purposes.

We used several methods to try to ensure that this national cohort is as complete as possible. We identified cases through "active negative surveillance"; that is, we actively sought cases from each center on a monthly basis and required centers to submit a "nil return" indicating that there were no cases so that we could be certain that all centers were reporting. We also used 2 additional sources of cases ascertainment: a national reporting system covering all consultant-led maternity units in the United Kingdom [9] and the British Isles Network of Congenital Anomalies Registers [10], which covers 50% of the births in the United Kingdom. Where cases were identified through either of these sources and not through BAPS-CASS, we contacted the appropriate pediatric surgeon and asked them to complete a data collection form. We are, therefore, confident that we have identified a high proportion of all affected infants, therefore allowing us to produce high quality generalizable information to inform the management of infants with gastroschisis in all countries with well-developed health care systems similar to that of the United Kingdom.

Table 3 Initial management for complex gastroschisis

Initial management, n (%) (n = 45)	Successful, n (%)	Converted, n (%)
Operative primary closure, 32 (71)	28 (86)	4 (14)
Ward reduction, 0 (0)	0 (0)	0 (0)
Preformed silo, 11 (24)	10 (91)	1 (9)
Custom silo, 1 (2)	1 (100)	0 (0)
Patch, 1 (2)	1 (100)	0 (0)

We divided cases into simple or complex gastroschisis according to the presence of additional bowel damage (perforation or atresia), as these factors are known to impact on choice of surgical technique as well as outcomes. Nearly 90% of infants had simple gastroschisis according to our classification. Many different general classifications may be used to stratify neonatal surgical patients according to their severity of illness, for example, SNAP-II and SNAPPE-II (Score for Neonatal Acute Physiology and SNAP Perinatal Extension) [11]. We chose to use a more straightforward classification of severity to limit the data collection burden for collaborating centers and to produce information that would be understandable and easily used for the purposes of counseling parents. The one other national prospective population-based study of the surgical management of gastroschisis, reporting on 100 infants with gastroschisis in Canada [12], did not attempt to separate the infants into these groups, possibly because the smaller number of cases severely limits the generalizability of any discussion of complex cases as a unique group. This classification has, however, been used in other retrospective national and hospital-based database studies, which report similar proportions [13,14].

Although this classification divides infants with gastroschisis into 2 broad groups, there are many other factors that

Table 4 Outcomes for simple and complex gastroschisis

	Simple, n (%) (n = 336)	Complex, n (%) (n = 45)	Risk ratio (95% CI)
Ventilated postoperatively			
No	76 (23)	3 (7)	1
Yes	260 (77)	42 (93)	1.21 (1.09-1.33)
Wound dehiscence			
No	320 (95)	42 (93)	1
Yes	16 (5)	3 (7)	1.40 (0.42-4.62)
Wound infection			
No	324 (97)	45 (100)	1
Yes	10 (3)	0 (0)	Not estimable
IFALD			
No	326 (97)	34 (76)	1
Yes	10 (3)	11 (24)	8.21 (3.70-18.2)
Abdominal compartment syndrome			
No	334 (99)	45 (100)	1
Yes	2 (<1)	0 (0)	Not estimable
Necrotizing enterocolitis			
No	304 (90)	39 (87)	1
Yes	32 (10)	6 (13)	1.40 (0.62-3.16)
Reoperation			
No	296 (88)	10 (22)	1
Yes	40 (12)	35 (78)	6.53 (4.70-9.09)
Neonatal death			
No	330 (98)	45 (100)	1
Yes	6 (2)	0 (0)	Not estimable
Received TPN [a] (d)			
<28	196 (60)	8 (18)	1
≥28	129 (40)	37 (82)	2.07 (1.71-2.51)

[a] Excludes infants who died.

influence the choice of surgical procedure, including factors relating to the infant, the surgeon, and the surgical unit, where it would be impossible to capture comprehensively in a study such as this. The only way that the outcomes from surgical procedures can be truly evaluated is when the characteristics of the groups being compared, both known and unknown, are entirely balanced, that is, in the context of a randomized controlled trial (RCT). Nevertheless, this national observational study does not demonstrate any clear benefit of one technique over another and thus provides important information to inform the development of an RCT.

The surgical strategy used most commonly by surgeons in the United Kingdom and Ireland was operative primary reduction and sutured fascial defect closure; the frequency of use was significantly different for simple and complex categories of gastroschisis (51% versus 71%). Staged reduction and delayed defect closure using preformed silos, most of which were placed nonoperatively at the bedside, were used for 36% and 24% of simple and complex cases, respectively, which was not significantly different. Nonoperative ward reduction was used exclusively for simple gastroschisis in keeping with the selection criteria suggested by Bianchi et al [6] in 2002 and only accounted for 6% of simple gastroschisis cases. A Cochrane review in 2002 found that the evidence neither "supports or refutes" ward reduction and recommended a RCT to address the question [15,16]. No trial has been undertaken to date, and the results of this national observational study suggest that the technique has not been widely adopted in the United Kingdom. There is a suggestion from these data that the technique is associated with a lower initial success rate than other techniques for

Table 5 Outcomes of simple gastroschisis according to initial management

	Operative fascial closure, n (%) (n = 170)	Preformed silo, n (%) (n = 120)	Risk ratio (95% CI)
Ventilated postoperatively			
No	21 (12)	44 (37)	1
Yes	149 (88)	76 (63)	1.38 (1.19-1.60)
Wound dehiscence			
No	166 (98)	111 (93)	1
Yes	4 (2)	9 (7)	0.31 (0.10-1.00)
Wound infection			
No	165 (97)	115 (96)	1
Yes	5 (3)	5 (4)	0.71 (0.21-2.38)
IFALD			
No	164 (96)	118 (98)	1
Yes	6 (4)	2 (2)	2.12 (0.43-10.3)
Abdominal compartment syndrome			
No	170 (100)	120 (100)	1
Yes	0 (0)	0 (0)	Not estimable
Necrotizing enterocolitis			
No	152 (89)	110 (92)	1
Yes	18 (11)	10 (8)	1.27 (0.61-2.65)
Reoperation			
No	146 (86)	110 (92)	1
Yes	24 (14)	10 (8)	1.69 (0.84-3.41)
Neonatal death			
No	167 (98)	118 (98)	1
Yes	3 (2)	2 (2)	1.06 (0.18-6.24)
Received TPN [a] (d)			
<28	109 (66)	54 (48)	1
≥28	56 (34)	59 (52)	0.65 (0.49-0.86)

[a] Excludes infants who died.

closure of simple gastroschisis, indicating that case selection criteria should be further developed before the decision to use this technique is made. It should be noted, however, that this higher conversion rate is entirely consistent with the intentions and suggestions of the original authors of these techniques, who advocate that in the first instance, appropriately selected patients should follow a nonoperative reduction with the option of immediate conversion in the event of any difficulties or concerns. This allows the option of nonoperative reduction for infants for whom it is suitable while reducing serious complication risks by allowing for early conversion if necessary.

In contrast, staged reduction and delayed defect closure using preformed silos has been widely adopted in the United Kingdom, being the intended initial technique in 33% of all cases. The potential advantages of nonoperative placement of preformed silos at the bedside soon after birth, whether followed by operative delayed defect closure or a nonoperative dressing closure, has been highlighted by retrospective studies and 1 RCT, which essentially would seem to be a reduced need for ventilatory support and general anesthesia, although outcomes in these studies were similar, and therefore, these benefits have not been proven [2,3]. The single previous population-based study by the Canadian Association of Pediatric Surgeons Network also used population-based data from 16 centers with similar success in data collection. They reported comparable outcomes between operative "urgent closure" and staged reduction with delayed defect closure using a preformed silo; however, as in our study, neither the surgical strategy nor the perinatal management was standardized [12].

Our data on defect closure is limited, but these national data seem to show a lower rate of prosthetic patch closure than some single-center studies [17], which suggests that visceroabdominal disproportion is largely overcome and closure techniques are mostly successful in bringing about fascial apposition. This, in view of the fact that only approximately half of all cases are managed with intended operative primary reduction and defect closure may indicate that visceroabdominal disproportion is largely correctly predicted, so avoiding abdominal compartment syndrome and that staged closure techniques are being appropriately used in these cases.

The conversion rate for all initial surgical strategies deployed for simple gastroschisis was 13%. For operative fascial closure, this was 18%, and for ward reduction, it was 28%, both of which rates seemed higher than the conversion rates for preformed silo (6%). Case characteristics clearly influence the surgeon's choice of initial surgical strategy and the subsequent success; thus, these rates cannot be validly compared statistically. However, the conversion rates importantly suggest that no single approach is universally applicable, and therefore, a rescue strategy is needed by all surgeons. It seems that a clinical pathway is emerging in which a nonoperative technique may be adopted with conversion to an operative strategy if difficulties are encountered. Formalizing this pathway may be a rational approach, and gaining a consensus for this would be helpful. Risk categorization has been used to describe cases, but applying this to inform decision making would require some support by the surgical community [13].

It would seem that the 4 main strategies used may be useful to surgeons from time to time to allow them to tailor their approach according to factors such as related bowel injury, visceroabdominal disproportion, associated anomalies, or complications such as necrotizing enterocolitis. An RCT, therefore, of operative primary reduction versus staged reduction using a preformed silo for simple gastroschisis is possibly warranted. An RCT of operative primary reduction versus nonoperative primary "ward" reduction would be more problematic because ward reduction has not been adopted widely. However, many proponents of preformed silos would argue that if there is no clear additional clinical benefit, then why perform emergency surgical closure that has greater resource utilization and increased out of hours activity, which could make recruitment of trial sites problematic. Furthermore, the expertise of using preformed silos requires some time to develop [8]. The occurrence of bowel ischemia and abdominal compartment syndrome remains a concern for all strategies, and vigilance in monitoring is essential. However, the results of this national observational study demonstrate no clear benefits of either technique and may thus go some way toward demonstrating the true clinical equipoise required for a trial to gain the support of the whole surgical community.

Consistent with previous reports [13,14,18], most registered cases were categorized as simple gastroschisis (85%), and infants with simple gastroschisis were statistically significantly less likely to be ventilated postoperatively, develop IFALD, undergo reoperation, or require TPN for longer than 4 weeks when compared with the complex group. Nonstatistically significant but potentially clinically significant increases in rates of wound infection and necrotizing enterocolitis were observed in the complex group. Interestingly, all the neonatal deaths occurred in the simple group, although this was not statistically significantly different because of the small numbers involved. It should also be noted that only 3 of these infants died from complications directly related to their gastroschisis. We recognize, however, that all the outcomes we studied are short-term outcomes, and we are continuing to follow these infants up further to 1 year of age to obtain data, in particular on mortality, TPN dependency, and organ transplantation.

The mortality of gastroschisis is expected to be trimodal, representing peaks related to 3 phases of care: antenatal, neonatal, and early infancy. We identified a neonatal mortality rate of 6 (2%) in the simple group which is similar to other studies [12] and which is usually attributed to associated anomalies or complications of initial surgical intervention. Although in less well-developed health care settings, this rate is much higher and is attributed to sepsis, shock, and intestinal ischemia [19], this study suggests that this mortality rate could still be reduced further in the

United Kingdom. The true overall mortality rate will include in utero death and termination, as identified from obstetric data sources, deaths related to complications of neonatal treatment, and a later peak in early infancy caused by IFALD and the complications of short bowel syndrome or adhesive small bowel obstruction. We hope to present these data after the completion of our 1-year follow-up study.

4. Conclusions

This study provides a comprehensive picture of current UK practice in the surgical management of gastroschisis. Furthermore, we have demonstrated the benefits of collaborative research to produce generalizable results as well as a national benchmark, which may be used for comparison by individual operators and centers. The population-based BAPS-CASS system allows for rapid case accrual, and we have clearly demonstrated better short-term outcomes for infants with simple gastroschisis compared with complex cases. The 1-year follow-up data will further help to elucidate prognostic factors and guide further research to optimize surgical management of gastroschisis as well as providing robust information to allow comprehensive parental counseling.

Acknowledgments

This study would not have been possible without the collaboration of BAPS-CASS reporting clinicians throughout the United Kingdom and Ireland.

Addenbrooke's Hospital	Adil Aslam, Christopher Parsons
Birmingham Children's Hospital	Anthony Lander, Andrew Robb
Bristol Royal Hospital For Children	Eleri Cusick
Chelsea and Westminster Healthcare Trust	Nicholas Madden, Clare Skerritt
Edinburgh Royal Hospital for Sick Children	Gordon MacKinlay, Boma Adikibi
Glasgow Royal Hospital for Sick Children	Atul Sabharwal
Great Ormond Street Hospital for Sick Children	Joe Curry
Hull Royal Infirmary	Sanja Besarovic
John Radcliffe Hospital	Paul Johnson, Javaid Sadiq
King's College Hospital	Mark Davenport
Leeds General Infirmary	Ian Sugarman, Anthony Owen
Leicester Royal Infirmary	Shawqui Nour
Norfolk and Norwich University Hospital	Thomas Tsang
Nottingham University Hospitals NHS Trust	Brian Davies, Richard Stewart
Our Lady's Hospital for Sick Children	Prem Puri, Farhan Tareen
Queen Charlotte's and Chelsea Hospital	Robin Abel
Royal Aberdeen Children's Hospital	Christopher Driver
Royal Alexandra Children's Hospital	Anies Mahomed
Royal Belfast Hospital for Sick Children	William A McCallion, Suzanne Lawther
Royal Liverpool Children's Hospital	Paul Losty, David Wilkinson
Royal London Hospital	Catherine Cord-Udy, Simon Phelps
Royal Manchester Children's Hospital	Antonino Morabito, Anju Goyal
Royal Victoria Infirmary	Gareth Hosie
Sheffield Children's Hospital	Sean Marven, Karen Lloyd
Southampton General	David Burge, Melanie Drewett
St George's Hospital	Bruce Okoye
University Hospital Lewisham	Catherine Richards
University Hospital of Wales	Simon Huddart, Daniela Vieten, Ram Shrestha

We would also particularly like to acknowledge Carole Harris, who administered the data collection for the study, and Judith Budd, Mary Bythell, Catherine Rounding, Rosie Thompson, Ann Tonks, David Tucker, and Diana Wellesley from the British Isles Network of Congenital Anomalies Registers, who helped with additional case ascertainment. The support of the British Association of Paediatric Surgeons contributed greatly to the success of the study.

References

[1] Bianchi A, Dickson AP. Elective delayed reduction and no anesthesia: 'minimal intervention management' for gastrochisis. J Pediatr Surg 1998;33:1338-40.
[2] Owen A, Marven S, Jackson L, et al. Experience of bedside preformed silo staged reduction and closure for gastroschisis. J Pediatr Surg 2006;41:1830-5.
[3] Pastor AC, Phillips JD, Fenton SJ, et al. Routine use of a SILASTIC spring–loaded silo for infants with gastroschisis: a multicenter randomized controlled trial. J Pediatr Surg 2008;43:1807-12.
[4] Caniano DA, Brokaw B, Ginn-Pease ME. An individualized approach to the management of gastroschisis. J Pediatr Surg 1990;25:297-300.
[5] Filston HC. Gastroschisis-primary fascial closure. The goal for optimal management. Ann Surg 1983;197:260-4.
[6] Bianchi A, Dickson AP, Alizai NK. Elective delayed midgut reduction—no anesthesia for gastroschisis: selection and conversion criteria. J Pediatr Surg 2002;37:1334-6.
[7] Donaldson L. On the state of the public health: annual report of the Chief Medical Officer 2004. London: Department of Health; 2005.

[8] Lansdale N, Hill R, Gull-Zamir S, et al. Staged reduction of gastroschisis using preformed silos: practicalities and problems. J Pediatr Surg 2009;44:2126-9.
[9] Knight M, Kurinczuk JJ, Tuffnell D, et al. The UK Obstetric Surveillance System for rare disorders of pregnancy. BJOG 2005;112:263-5.
[10] BINOCAR: British Isles Network of Congenital Anomaly Registers. Available at: http://www.binocar.org/index.htm. Accessed August 2009.
[11] Richardson DK, Corcoran JD, Escobar GJ, et al. SNAP-II and SNAPPE-II: simplified newborn illness severity and mortality risk scores. J Pediatr 2001;138:92-100.
[12] Skarsgard ED, Claydon J, Bouchard S, et al. Canadian Pediatric Surgical Network: a population-based pediatric surgery network and database for analyzing surgical birth defects. The first 100 cases of gastroschisis. J Pediatr Surg 2008;43:30-4.
[13] Arnold MA, Chang DC, Nabaweesi R, et al. Risk stratification of 4344 patients with gastroschisis into simple and complex categories. J Pediatr Surg 2007;42:1520-5.
[14] Payne NR, Pfleghaar K, Assel B, et al. Predicting the outcome of newborns with gastroschisis. J Pediatr Surg 2009; 44:918-23.
[15] Davies MW, Kimble RM, Woodgate PG. Ward reduction without general anaesthesia versus reduction and repair under general anaesthesia for gastroschisis in newborn infants. Cochrane Database Syst Rev 2002:CD003671.
[16] Davies MW, Kimble RM, Cartwright DW. Gastroschisis: ward reduction compared with traditional reduction under general anesthesia. J Pediatr Surg 2005;40:523-7.
[17] Saxena AK, Hulskamp G, Schleef J, et al. Gastroschisis: a 15-year, single-center experience. Pediatr Surg Int 2002;18:420-4.
[18] Keys C, Drewett M, Burge DM. Gastroschisis: the cost of an epidemic. J Pediatr Surg 2008;43:654-7.
[19] Sangkhathat S, Patrapinyokul S, Chiengkriwate P, et al. Infectious complications in infants with gastroschisis: an 11-year review from a referral hospital in southern Thailand. J Pediatr Surg 2008;43: 473-8.