Evolution of management of gastroschisis

Amel Abd Eltwab Hashish and Essam Elhalaby

Background/purpose **The management protocols and outcome of neonates with gastroschisis have improved significantly during the past two decades. The purpose of this study was to evaluate the evolution in management and outcome of gastroschisis in our institution.**

Materials and methods **All patients treated for gastroschisis during the past 12 years were included. These patients were divided into two chronologically distinct groups. Group I included patients who were treated from 1998 to 2005 and group II included patients who were treated from 2005 to 2010. Each group was further subdivided into two subgroups according to the method of closure of the abdominal wall by either primary (group IA and group IIA) or delayed primary closure of the abdominal wall defect after temporary extra-abdominal hosting of the bowel using hand-sewn silastic or plastic sheets (group IB) or a spring-loaded silo (group IIB). Each patient was evaluated with regard to time spent on ventilator, time to initiating enteral feeds, time to discharge from the Neonatal Intensive Care Unit, and any complications.**

Results **There was no difference between the two main groups with regard to the gestational age, sex, mode of delivery, or the percentage of associated congenital anomalies. Primary closure was feasible in 29 patients (18 in group IA and 11 in group IIA). Staged reduction of the herniated bowel and delayed repair were performed in 23 patients (12 in group IB and 11 in group IIB). Reduction of the herniated bowel and delayed staged reduction were performed earlier in group IIB than in group IB. Enteral feeding was earlier in patients who had primary closure either in group IA or group IIA compared with patients treated with delayed closure in either group IB or group IIB. Enteral feeding was relatively earlier in group IIB than in group IB, but the difference was not significant.**

Conclusion **The overall morbidity and mortality showed significant improvement in the management of gastroschisis at our practice. The introduction of spring-loaded silo has simplified the management of patients born with gastroschisis who cannot be treated with primary reduction. Primary closure continued to have better outcome measures compared with staged closure.** ***Ann Pediatr Surg*** **7:10–15 © 2011 Annals of Pediatric Surgery**

Annals of Pediatric Surgery 2011, **7:**10–15

Keywords: abdominal wall defect, complications, gastroschisis spring-loaded silo

Department of Pediatric Surgery, Tanta University, Tanta, Egypt

Correspondence to Amel Abd Eltwab Hashish, Department of Pediatric Surgery,Tanta University Hospital, Elgish Street, Tanta, 31111, Egypt
Tel: 0020 40 333 5695, 20 12 331 5309; fax: 00 20 40 341 2127;
e-mail:amelhashish@gmail.com

Received 12 August 2010 **Accepted** 25 September 2010

Introduction

Gastroschisis is a full thickness defect of the abdominal wall with herniation of a variable amount of uncovered intestinal loops through a defect immediately to the right of a normally formed umbilicus. The intestine is frequently foreshortened, covered with gelatinous exudates, matted together, and/or is edematous due to its exposure to amniotic fluid and compression of the mesenteric blood supply at the defect. The incidence of gastroschisis is rising over the last decade. It has been estimated between one in 3000 and one in 10 000 live births [1–3].

The management of gastroschisis has gradually evolved and improved over the years. The principles of management however remain the same. First, to reduce the viscera safely, second, to close the abdominal wall defect with an acceptable cosmetic appearance and, third, proper nutrition support, in addition to detection and proper management of any associated anomalies or complications [4].

The first surgical intervention by manual closure with fatal outcome was reported in 1878 [5]. In 1943, Watkins [6] reported the first successful primary closure of gastroschisis. In 1953, Moor and Stokes [7] reported the use of skin flap technique to close the abdomen. However, two of their patients died because of acute respiratory insufficiency and abdominal compartmental syndrome.

Historically, many surgeons used drastic measures to decrease the size of the abdominal contents and to relieve size discrepancy, such as bowel resection, splenectomy, and partial hepatectomy [8,9]. In 1966, manual stretching of the abdominal wall was advocated by Izant *et al.* [10] to enlarge the abdominal cavity. However, the overzealous stretching was complicated by lateral abdominal wall hernia [11].

The real advance in surgical management of gastroschisis was the introduction of staged reduction by using sheets of Teflon as a temporary covering for the herniated bowel by Schuster in 1967 [12]. However, the abdomen required periodic reopening to perform serial reduction and staged removal of the material with risk of bowel injury and infectious complications. The modification of Schuster's technique by Allen and Wrenn [13] involved the use of silastic instead of Teflon, with no attempt at skin coverage to facilitate serial reduction and enable continuous inspection of the bowel's condition.

In 1975, Shermata and Haller [14] used a preformed transparent silo sutured to the abdominal wall. The silo was

1687-4137 © 2011 Annals of Pediatric Surgery DOI: 10.1097/01.XPS.0000393094.25977.48

suspended to allow relief of bowel edema and also to allow a gradual reduction of the viscera into the abdominal cavity.

Starting from 1997, surgeons began inserting a silastic silo (Dow Corning, Midland, Michigan, USA) or a spring-loaded silo (SLS, Bentec, Sacramento, California, USA) when primary closure was not feasible. SLS could be placed at bedside and it alleviated the need for suturing to the abdominal wall [15]. Minkes *et al.* [16] reported that gradual reduction using SLS seems to eliminate the need for intraoperative pressure measurement.

The aim of this study was to evaluate the progress in the management and outcome of cases of gastroschisis at the Tanta University Hospital during the past decade.

Materials and methods

Patients were divided into two chronologically distinct groups. Group I (from 1998 to 2005) included 30 patients and group II (from 2005 to 2010) included 22 patients. Each group was further subdivided into two subgroups according to the method of closure of the abdominal wall by either primary (group IA and group IIA) or delayed primary closure of the abdominal wall defect after temporary extra-abdominal hosting of the bowel using silastic or plastic sheets (group IB) or SLS (group IIB).

Initial management aimed at maintaining circulation to the bowel and preventing infection by covering the defect with sterile dressing soaked in warm saline to prevent fluid loss, stabilizing infant (temperature/fluids), gastric decompression, intravenous fluids with glucose, and broad-spectrum antibiotics. After stabilization of the infants in the Neonatal Intensive Care Unit, the repair was performed primarily whenever feasible after reducing the herniated bowel (Fig. 1a and b). The decision of primary closure was based on the size of the herniated viscera and size of the abdominal cavity.

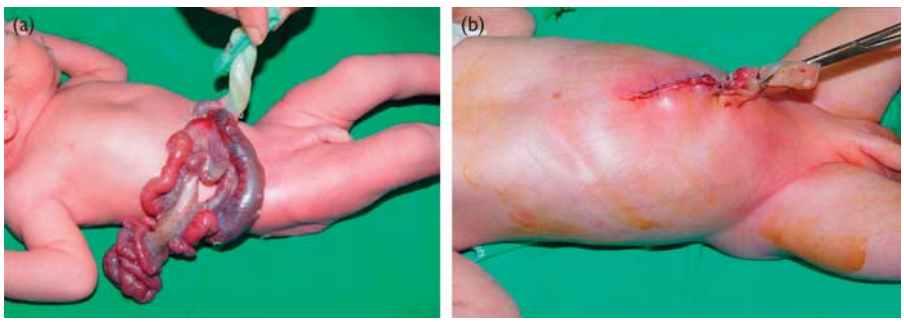
When primary closure was not possible due to the presence of discrepancy between the size of the herniated bowel and the volume of abdominal cavity, a staged reduction of the herniated bowel and delayed closure of the abdomen was performed after temporary extra-abdominal hosting of the bowel in a chimney made of a silastic sheet or any other available plastic sheet, when silastic sheet was not available, (Figs. 2 and 3) (group IB). The sheet was sewn directly to the fascia after mobilizing skin flaps circumferentially around the defect. Postoperatively, running stitches through the silastic/plastic sheet were made to initiate gradual reduction of the extruded intestine over 8–15 days till the abdominal wall got lax and the abdominal cavity could contain the extruded part after which the defect was closed [13].

Staged reduction and delayed primary repair were performed in group IIB patients using a SLS (Bentec), which was placed over the exposed viscera under the fascial defect using complete aseptic precautions (Figs. 4 and 5). The SLS was inserted either at Neonatal Intensive Care Unit (five cases) or in the operating room (six patients). Adhesions from the fascia to the bowel wall were gently disrupted manually. In cases of dense adhesions to the bowel wall or very small opening, silo placement was performed in the operating room to allow lyses of adhesions or widening of the fascial defect. The bowel was gradually reduced over the next few days. The transparent materials of the silo allow for continuous inspection of the bowel. No sutures were used. The base of the silo was wrapped with gauze soaked in antiseptic solution. The silo was closed with umbilical tape over the next few days and the abdominal viscera were reduced progressively. When the bowel was completely reduced, a second stage closure was performed in the operating room [15].

Fig. 2

Hand-sewn bag (group 1B).

Fig. 1



(a) Noncomplicated gastroschisis. (b) Primary closure 1 h after delivery.

Each patient was evaluated with regard to time to full closure of the abdominal wall, time spent on ventilator if needed, time to initiating enteral feeds, time to discharge from the Neonatal Intensive Care Unit, and any complications.

Statistical analysis

Statistical analysis was carried out using SPSS ver. 13 (Chicago, Illinois, USA). All quantitive data were compared using the student *t* test, whereas all qualitative data were compared using the Fisher's exact test. A *P* value of less than 0.05 was considered to be significant.

Fig. 3

Hand-sewn silo (group 1B).

Results

Fifty-two neonates (14 male and 16 female) with gastroschisis were treated at the Tanta University Hospital and affiliated hospitals from June 1998 to October 2010.

Prenatal diagnosis was known in 13 patients (25%). The mean birth weight was 2.4 ± 0.89 kg. The median maternal age was 27 years (range: 19.5–41 years). Thirty-five patients had normal vaginal delivery. Associated congenital anomalies were present in 10 patients (six in group I and four in group II). There was no difference between the two main groups with regard to the gestational age (36.9 ± 2.3 vs. 36.5 ± 2.5 weeks), sex, mode of delivery, or the percentage of associated congenital anomalies.

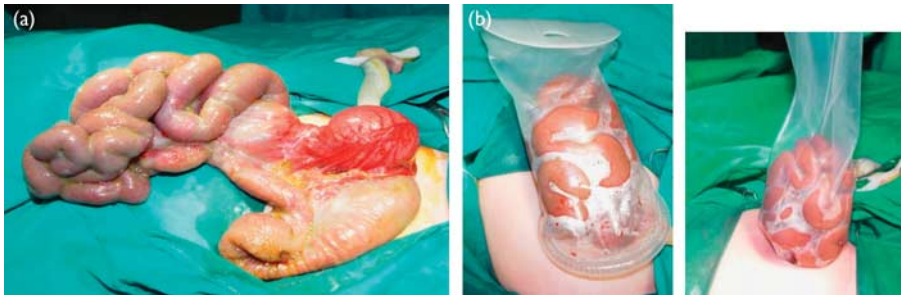
Prenatal diagnosis of abdominal wall defect was noted in five (16.7%) patients in group I and in seven (31.8%) patients in group II (difference not statistically significant, $P = 0.35$). Five patients had associated intestinal atresia (Fig. 6). Primary anastomosis was performed in two cases with little edematous bowel. Ileostomy was performed in two other cases. One patient died before definitive surgery of the associated intestinal atresia; her bowel was placed into the abdomen for nasogastric decompression planning for reoperation.

Primary closure was feasible in 29 patients (18 in group IA, and 11 in group IIA). Staged reduction of the

Fig. 4

(a) Gastroschisis: foreshortened, edematous bowel. (b) Staged reduction using spring-loaded silo.

Fig. 5



(a) Noncomplicated gastroschisis. (b) Staged reduction using spring-loaded silo.

herniated bowel and delayed repair were needed in 23 patients (12 in group IB, and 11 in group IIB). Primary closure was performed 30 min–10 h after delivery. The mean time from birth to primary closure was shorter in the second group compared with the first group (5 ± 1 vs. 7 ± 2 h, the difference is statistically significant, $P = 0.004$).

Reduction of the herniated bowel and delayed staged reduction were earlier in group IIB than in group IB (mean: 8 ± 2 vs. 11 ± 3 days, the difference is statistically significant, $P = 0.01$). Enteral feeding was earlier in patients who had primary closure either in group IA or group IIA compared with patients treated with delayed closure in either group IB or group IIB. The mean time of first oral feeding was earlier in patients who had primary closure either in group I or group II compared with patients undergoing delayed closure. Enteral feeding was relatively earlier in group IIB than in group IB and, but the difference was not statistically significant ($P = 0.36$).

The duration of hospital stay was significantly longer for patients who had staged surgery either in group I or group II than those who had primary closure (total mean: 12 ± 4 vs. 18 ± 3 days, $P = 0.0001$) (Table 1).

The various postoperative complications are summarized in Table 2. Seven deaths occurred in group I due to infection and septicemia in three patients and prolonged bowel dysmotility in another four patients. Two patients died in group II: one due to sepsis and cardiac anomalies and another due to ischemia of the foregut because of vascular insufficiency at the level of the abdominal wall, leading to significant intestinal loss (Fig. 7). The frequency of mortality (23 vs. 9%) improved steadily over the years.

Repair of postclosure of incisional hernia was needed in seven patients in group I and in three patients in group II.

Follow-up period ranged from 2 to 6 months in both groups (average: 3.7 months). Two patients came later with adhesive intestinal obstruction that was treated conservatively and another one with inguinal hernia.

Fig. 6

Gastroschisis associated with intestinal atresia and bowel perforation.

Discussion

Gastroschisis continued to be a challenging surgical emergency in neonates. The abdominal cavity may not be large enough to tolerate reduction of herniated frequently foreshortened, edematous, matted together bowel in many cases. Similarly, a prolonged ileus represents a challenge in the postoperative period.

During the past 12 years, there was a steady improvement in antenatal care at our region. Prenatal diagnosis of abdominal wall defect increased from 16.7% in group I to 31.8% in group II. All mothers were advised to continue the normal course of pregnancy instead of preterm delivery. The limited incubators equipped with advanced neonatal intensive care facilities dictated this approach.

Preterm delivery of infants with gastroschisis was recommended by some researchers to avoid the intestinal damage that may occur due to prolonged exposure to the amniotic fluid, which contain inflammatory factors that lead to intestinal ischemia or damage [17,18]. In contrary, Maramreddy *et al.* [19] reported that there is no benefit of preterm delivery in reducing the morbidities in patients with gastroschisis. In addition, preterm delivery increased complication rate in those infants with regard to sepsis, longer hospital stay, and prolonged period to establish feeding and to tolerate full feeding.

Owing to increased prenatal diagnosis, more deliveries were performed at hospitals equipped with neonatal care facilities. Earlier presentation and primary closure was noted in group II compared with group I (5 vs. 7 h). This might have contributed to the overall improvement of outcome in the second group. Stringer *et al.* [20] reported that prenatal diagnosis and transfer of patient to a specialized center is in favor of more frequent successful primary closure, less postoperative ventilation, and reduced hospital stay, because of earlier surgery and early

Table 1 **Patient characteristics**

	Group I		Group II	
	Primary ($n=18$)	Staged closure ($n=12$)	Primary ($n=11$)	Spring-loaded silo ($n=11$)
Prenatal diagnosis	2	3	3	4
Time at closure (mean)	7 ± 2 h	11 ± 3 days	5 ± 1 h	8 ± 2 days
Patients requiring ventilation	4	2	1	1
Time to start enteral feeding (mean in days)	7 ± 2	11 ± 3	6 ± 2	12 ± 2
Time to full feeding (range in days)	7 – 23	15 – 30	7 – 19	13 – 20
Days of hospitalization (mean)	12 ± 3	20 ± 2	11 ± 2	17 ± 1
Associated intestinal atresia	2	1	1	1

Table 2 Postoperative complications in 52 patients with gastroschisis

	Group I		Group II	
	Primary ($n=18$)	Staged closure ($n=12$)	Primary ($n=11$)	Spring-loaded silo ($n=11$)
Partial wound dehiscence	4	4	1	1
Complete wound disruption	1	2	0	0
Incisional hernia	4	3	2	1
Wound infection	2	4	1	1
Intestinal obstruction	1	0	1	0
Mortality	4	3	1	1

Fig. 7

Gastroschisis complicated with massive intestinal ischemia.

management of patient with regard to hypothermia and hypovolemia and adequate nasogastric drainage.

The management of neonates with gastroschisis depends on several factors including the amount and status of herniated bowel, the size of abdominal cavity, the available resources at neonatal intestine care unit, and the presence or absence of other associated congenital anomalies. Operative primary reduction with closure of the abdominal defect continued to be 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 [4].

A significant percentage of infants with gastroschisis can undergo reduction of the herniated intestinal contents and primary abdominal wall closure. Primary closure was decided whenever feasible. In 1970, pharmacologic paralysis and prolonged mechanical ventilation after aggressive attempts at primary closure were recommended [21].

More patients underwent primary repair in the first group (60% of cases). For fear of infection and disruption of the silo at the margin of the defect and risk of evisceration, many surgeons still prefer primary closure in many cases of gastroschisis, if at all possible. The use of intraoperative pressure manometry by measuring either intragastric central venous pressure or bladder was recommended as a guide for primary closure of the abdominal defect to avoid potential abdominal compartment syndrome, which may lead to ischemic/necrotic bowel, renal insufficiency, and respiratory distress [22–24].

Operative staged reduction has been predominantly achieved by suturing a synthetic material to the enlarged defect and delayed defect closure. Staged closure carries the risk of loss of fascial strength at the margins of the defect, the infection risk from the lack of a watertight seal, and the risk of evisceration due to disrupt suture line [25].

A more conservative approach toward overzealous primary closure has been adopted in our practice during the past 5 years. The availability of SLS has encouraged us to limit primary closure to cases in which this can be done safely without the need of postoperative ventilation. The use of a preformed SLS bag in infants with gastroschisis has been shown to be associated with improved facial closure rates, fewer ventilator days, more rapid return of bowel function, and fewer complications. Reduction of gastroschisis bowel has been successfully performed with and without anesthesia [26,27].

Similar to other earlier published studies [16], we found that the length of hospital stay was longer in patients who had hand-sewn silo pouch (group IB) in comparison with those who had SLS (group IIB). Some studies highlighted the advantages of SLS such as: early and simple placement in the Neonatal Intensive Care Unit, allowing bowel reduction to occur under direct vision at an appropriate rate for each patient and allowing elective closure of the final defect, in addition to direct observation of the bowel for signs of ischemia and removal of the umbilical tapes if the bowel becomes ischemic or if ventilation and perfusion become compromised [26–30].

Despite the several advantages of SLS, it has potential pitfalls including ischemic complications, dislodgment, bowel twisting, and difficulties with final closure. One of the disadvantages of SLS is the progressively increased abdominal wall defect, which may be explained by the development of lateral distractive forces being applied to the abdominal wall [31]. Another potential complication is bowel ischemia at the inferior portion of the silo during reduction through small openings that can serve as a constriction point of the bowel or mesentery 'funnel effect' [32].

Many patients with gastroschisis have associated intestinal abnormalities including atresia (mostly involving the small intestine) [4,33]. The incidence of associated

atresia was 9.6% in this series. Although primary anastomosis is not recommended in such cases, this was feasible in two cases with relatively mild edematous bowel.

The outcome of patients with gastroschisis has dramatically improved in our institution and affiliated hospitals. The mortality rate was 23% during the first 7 years in this study, whereas it came down to less than 10% in the past 5 years. The improvement in outcome is related to the optimization of perinatal care, the availability of intravenous nutrition, and the use of staged closure when indicated. Late complications and mortality are related to sepsis either from an intra-abdominal or wound complication or from a central venous catheter placed for parental nutrition.

Conclusion

Significant changes occurred in the management philosophy of gastroschisis at our institution. The morbidity and mortality rates are steadily improving. Antenatal diagnosis is increasing. Primary closure of the abdominal wall defect is only performed if it is safe without high intra-abdominal pressure. The use of a SLS is helpful but has potential pitfalls.

References

1 Caniano DA, Brokaw B, Ginn Pease ME. An individualized approach to the management of gastroschisis. *J Pediatr Surg* 1990; **25**:297–300.
2 Baerg J, Kaban G, Tonita J, Pahwa P, Reid D. Gastroschisis: a sixteen-year review. *J Pediatr Surg* 2003; **38**:771–774.
3 Hougland KT, Hanna AM, Meyers R, Null D. Increasing prevalence of gastroschisis in Utah. *J Pediatr Surg* 2005; **40**:535–540.
4 Owen A, Marven S, Johnson P, Kurinczuk J, Spark P, Draper ES, *et al.* Gastroschisis: a national cohort study to describe contemporary surgical strategies and outcomes. *J Pediatr Surg* 2010; **45**:1808–1816.
5 Fear W. Congenital extrusion of abdominal viscera: return: recovery. *Br Med J* 1878; **2**:518–520.
6 Watkins DE. Gastroschisis. *Va Med* 1943; **70**:42–45.
7 Moore TC, Stokes GE. Gastroschisis. Report of two cases treated by a modification of the gross operation for omphalocele. *Surgery* 1953; **33**:112–120.
8 Buchanan RW, Cain WL. A case of a complete omphalocele. *Ann Surg* 1956; **143**:552–556.
9 Kleinhaus S, Kaufer N, Boley SJ. Partial hepatectomy in omphalocele repair. *Surgery* 1968; **64**:484–485.
10 Izant RJ Jr, Brown F, Rothmann BF. Current embryology and treatment of gastroschisis and omphalocele. *Arch Surg* 1966; **93**:49–53.
11 Firor HV. Technical improvements in the management of omphalocele and gastroschisis. *Surg Clin North Am* 1975; **55**:129–134.
12 Schuster SR. A new method for the staged repair of large omphaloceles. *Surg Gynecol Obstet* 1967; **125**:837–850.
13 Allen RG, Wrenn EL Jr. Silon as a sac in the treatment of omphalocele and gastroschisis. *J Pediatr Surg* 1969; **4**:3–8.
14 Shermeta DW, Haller JA Jr. A new preformed transparent silo for the management of gastroschisis. *J Pediatr Surg* 1975; **10**:973–975.
15 Kidd JN, Levy MS, Wagner CW. Staged reduction of gastroschisis: a simple method. *Pediatr Surg Int* 2001; **17**:242–244.
16 Minkes RK, Langer JC, Mazziotti MV, Skinner MA, Foglia RP. Routine insertion of a silastic spring-loaded silo for infants with gastroschisis. *J Pediatr Surg* 2000; **35**:843–846.
17 Guibourdenche J, Berrebi D, Vuillard E, De Lagausie P, Aigrain Y, Oury JF, *et al.* Biochemical investigations of bowel inflammation in gastroschisis. *Pediatr Res* 2006; **60**:565–568.
18 Hadidi A, Subotic U, Goeppl M, Waag KL. Early elective cesarean delivery before 36 weeks versus late spontaneous delivery in infants with gastroschisis. *J Pediatr Surg* 2008; **43**:1342–1346.
19 Maramreddy H, Fisher J, Slim M, LaGamma EF, Parvez B. Delivery of gastroschisis patients before 37 weeks of gestation is associated with increased morbidities. *J Pediatr Surg* 2009; **44**:1360–1366.
20 Stringer MD, Brereton RJ, Wright VM. Controversies in the management of gastroschisis: a study of 40 patients. *Arch Dis Child* 1991; **66** (**1 Spec No**):34–36.
21 Raffensperger JG, Jona JZ. Gastroschisis. *Surg Gynecol Obstet* 1974; **138**:230–234.
22 Wesley JR, Drongowski R, Coran AG. Intragastric pressure measurement: a guide for reduction and closure of the silastic chimney in omphalocele and gastroschisis. *J Pediatr Surg* 1981; **16**:264–270.
23 Lacey SR, Carris LA, Beyer AJ III, Azizkhan RG. Bladder pressure monitoring significantly enhances care of infants with abdominal wall defects: a prospective clinical study. *J Pediatr Surg* 1993; **28**:1370–1374; discussion 1374–1375.
24 Yaster M, Scherer TL, Stone MM, Maxwell LG, Schleien CL, Wetzel RC, *et al.* Prediction of successful primary closure of congenital abdominal wall defects using intraoperative measurements. *J Pediatr Surg* 1989; **24**:1217–1220.
25 Stringel G. Large gastroschisis: primary repair with Gore-Tex patch. *J Pediatr Surg* 1993; **28**:653–655.
26 David AL, Tan A, Curry J. Gastroschisis: sonographic diagnosis, associations, management and outcome. *Prenat Diagn* 2008; **28**:633–644.
27 Owen A, Marven S, Jackson L, Antao B, Roberts J, Walker J, *et al.* Experience of bedside preformed silo staged reduction and closure for gastroschisis. *J Pediatr Surg* 2006; **41**:1830–1835.
28 Wu Y, Vogel AM, Sailhamer EA, Somme S, Santore MJ, Chwals WJ, *et al.* Primary insertion of a silastic spring-loaded silo for gastroschisis. *Am Surg* 2003; **69**:1083–1086.
29 Allotey J, Davenport M, Njere I, Charlesworth P, Greenough A, Ade Ajayi N, *et al.* Benefit of preformed silos in the management of gastroschisis. *Pediatr Surg Int* 2007; **23**:1065–1069.
30 Pastor AC, Phillips JD, Fenton SJ, Meyers RL, Lamm AW, Raval MV, *et al.* Routine use of a SILASTIC spring-loaded silo for infants with gastroschisis: a multicenter randomized controlled trial. *J Pediatr Surg* 2008; **43**:1807–1812.
31 Lobo JD, Kim AC, Davis RP, Segura BJ, Alpert H, Teitelbaum DH, *et al.* No free ride? The hidden costs of delayed operative management using a spring-loaded silo for gastroschisis. *J Pediatr Surg* 2010; **45**:1426–1432.
32 Ryckman J, Aspirot A, Laberge JM, Shaw K. Intestinal venous congestion as a complication of elective silo placement for gastroschisis. *Semin Pediatr Surg* 2009; **18**:109–112.
33 Arnold MA, Chang DC, Nabaweesi R, Colombani PM, Bathurst MA, Mon KS, *et al.* Risk stratification of 4344 patients with gastroschisis into simple and complex categories. *J Pediatr Surg* 2007; **42**:1520–1525.