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Gastroschisis with intestinal atresia—predictive value of antenatal diagnosis and outcome of postnatal treatment☆

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Abstract

Purpose: The purpose of this study is to evaluate (1) the predictive value of fetal bowel dilatation (FBD) for intestinal atresia in gastroschisis and (2) the postnatal management and outcome of this condition.

Methods: A retrospective review of all gastroschisis cases diagnosed in our fetal medicine unit between 1992 and 2010 and treated postnatally in our center was performed.

Results: One hundred thirty cases had full postnatal data available. Intestinal atresia was found at surgery in 14 neonates (jejunum, n = 6; ileum, n = 3; ascending colon, n = 3; multiple, n = 2). Polyhydramnios and FBD were more likely in the atresia group compared with infants with no atresia ($P = .0003$ and $P = .005$, respectively). Fetal bowel dilatation had 99% negative predictive value (95% confidence interval, 0.9-0.99) and 17% positive predictive value (95% confidence interval, 0.1-0.3) for atresia. Treatment of intestinal atresia included primary anastomosis (n = 5), delayed anastomosis (n = 2), and stoma formation followed by anastomosis (n = 7). Infants with atresia had longer duration of parenteral nutrition, higher incidence of sepsis, and cholestasis compared with infants with no atresia ($P = .0003$). However, the presence of atresia did not increase mortality.

Conclusions: Polyhydramnios and FBD are associated with atresia. Absence of FBD in gastroschisis excludes intestinal atresia. In our experience, atresia is associated with a longer duration of parenteral nutrition but does not influence mortality. These findings may be relevant for antenatal counseling.
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Gastroschisis is characterized by a full-thickness abdominal wall defect associated with herniation of the gut and other organs and has an incidence of up to 5 of 10,000 live births [1]. Other abnormalities may be seen in approximately 15% of infants, involving the central nervous system, heart, musculoskeletal system, and kidneys [2,3], and these have been associated with poor outcome. However, gastroschisis is an isolated anomaly isolated in most infants, and the outcome is influenced by

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intestinal dysmotility and malabsorption [4-8]. Survival rates for uncomplicated, isolated gastroschisis currently exceed 90% [7,9-12]. For infants with additional bowel pathology, long-term survival is often complicated by intestinal failure, parenteral nutrition (PN)–associated cholestasis, and sepsis with a reported mortality of almost 50% [13]. In 10% to 20% of cases, gastroschisis is associated with intestinal, usually small bowel, atresia [9,14-16], and several reports have shown this to have increased morbidity and mortality [17-22].

Routine maternal sonography ensures that almost all cases of fetal gastroschisis are diagnosed antenatally [23]. The aim of such assessment is to examine for associated abnormalities that may be associated with an adverse outcome and to detect indicators of fetal compromise leading to complications, which might be averted by prompt delivery [1]. There are conflicting findings on the usefulness of prenatal bowel abnormalities to predict bowel pathology in gastroschisis, in particular for the prognostic significance of fetal bowel dilatation (FBD). This has been associated with a worse clinical outcome [24,25], a significantly longer time to full oral feeding and a greater need for bowel resection [26], and with complications such as bowel obstruction or atresia, necrosis, and need for bowel resection [20]. One large study proposed that FBD in the second trimester predicted neonatal bowel atresia [27]. However, another study found that similar findings had no correlation with postnatal outcome [28].

The aim of this study was to evaluate (1) the predictive value of antenatally detected bowel dilatation for atresia and (2) the postnatal management and outcome of infants with gastroschisis associated with intestinal atresia.

1. Materials and methods

Retrospective study of all live-born infants with antenatally diagnosed gastroschisis treated in our institution from January 1992 to December 2010. Ethical approval for the study was obtained (study 09SG12).

All women referred to the Fetal Medicine Unit at University College London Hospital (UCLH) with pregnancies affected by gastroschisis were identified using the fetal database (Viewpoint; General Electric, Wauwatosa, WI). Only those women whose baby was delivered at UCLH and who underwent surgery at UCLH (before 2000) or Great Ormond Street Hospital for Children (after 2000) were included in the study.

Of the 161 cases of gastroschisis initially referred to the Fetal Medicine Unit, 5 women requested termination of pregnancy, and there was 1 misdiagnosis of a ruptured omphalocele. Of the remaining 155 ongoing pregnancies, 18 women transferred their antenatal care to deliver at other units and were excluded from analysis. One hundred thirty-seven women continued with their pregnancy and delivered at our institution; 1 woman had a late miscarriage (21^6 weeks). Postnatal records were incomplete in 6 cases, leaving 130 for full analysis.

Details of sonographic examination and fetal images were entered onto the database at the time of the ultrasound examination. Women were scanned every 4 weeks until 30 weeks and then every 2 weeks thereafter or weekly if fetal compromise was suspected. At every fetal scan, the maximum bowel diameter in the intra-abdominal and extra-abdominal bowel was measured using electronic calipers placed on opposite inner bowel walls. Fetal bowel dilatation (FBD) was diagnosed when the bowel diameter was 18 mm or more. Various cutoffs for bowel dilatation have been applied over the years in the literature. Management of fetal gastroschisis at UCLH has been based on a cutoff of 18 mm or more [29]. This was first suggested in 1993 by Langer et al [26] and has been supported by later studies [24,30,31].

Our management included scheduled induction of labor at between 37 and 38 weeks of gestation, unless there was evidence of poor fetal growth velocity in association with abnormal umbilical artery Doppler examination or sudden onset of polyhydramnios, in which case earlier delivery was arranged. Mode of delivery was determined by obstetric considerations; planned cesarean delivery was performed for maternal reasons or where there was evidence of fetal compromise or in the case of a very large hernial defect.

Neonatal records were examined to ascertain outcome characteristics including gestational age at delivery, birth weight, Apgar score, and the presence of further structural anomalies. At delivery, immediate management of the neonate included assessment of vital signs, passage of a nasogastric tube to decompress the stomach, and establishment of intravenous access. The eviscerated bowel was protected by wrapping the infant's abdomen with cling-film, ensuring that the bowel was maintained in the midline to prevent kinking at the abdominal defect and resultant interruption of the mesenteric blood supply. Intravenous broad spectrum antibiotics were commenced, and the neonate was transferred for surgery. The surgical approach, the number and type of surgical interventions, duration of PN, duration of hospitalization, complications, and long-term outcomes were recorded.

Intestinal failure was defined as PN dependency for 28 days or more as defined by the British Intestinal Failure Survey [32]. Sepsis was diagnosed based on a positive culture in a blood sample derived from the central line. Parenteral nutrition–associated cholestasis was diagnosed when serum levels of conjugated bilirubin measured greater than 2.0 mg/dL in at least 2 repeated samples [33].

1.1. Statistical analysis

Statistical analysis was carried out using SPSS (SPSS, Chicago, IL), Graph Pad Prism 4 (GraphPad Software Inc, San Diego, CA), and VasserStat (http://faculty.vassar.edu/lowry/VassarStats.html). Data were summarized using the

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