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Gastroschisis associated with bladder evisceration complicated by hydronephrosis presenting antenatally

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ABSTRACT

We report here a case of gastroschisis associated with bladder evisceration and complicated by rapidly developing hydronephrosis diagnosed antenatally. The timing of delivery was determined by the hydronephrosis, associated bowel dilatation and polyhydramnios. The case highlights the need for continuing ultrasonographic surveillance of fetuses with gastroschisis to identify further associated complications which were hitherto absent but whose presence may influence the timing of delivery and neonatal care.

INTRODUCTION

The incidence of gastroschisis is reported to be rising[1,2]. In this condition, the commonest organs to eviscerate are the bowel and stomach. We report a rare case of gastroschisis associated with bladder evisceration, polyhydramnios and hydronephrosis.

CASE REPORT

A 25-year-old woman, gravida 4, para 1+2 (normal spontaneous vaginal delivery at 39 weeks, weight 3.17 kg), a warehouse worker who smoked, was referred from a district general hospital for tertiary assessment and management at Leicester following a diagnosis of gastroschisis made at 16 weeks' gestation. A routine anomaly ultrasound scan at 19 weeks' gestation had excluded other abnormalities. The pregnancy had so far been uncomplicated.

Further ultrasonographic assessment at the Leicester Royal Infirmary at 24 weeks' gestation confirmed the diagnosis and the absence of other structural abnormalities. The fetal growth was satisfactory and the amniotic fluid volume was normal. Four weekly ultrasound scans were planned after the woman had been seen by the pediatric surgeon and the neonatologist. At 28 weeks' gestation, fetal growth was again normal and there was no polyhydramnios. In view of the fact that the patient lived 44 miles from Leicester, elective delivery was planned for 38 weeks' gestation. However, at follow-up scanning at 34 weeks' gestation, there was polyhydramnios (maximum pool depth 9.3 cm), a distended bladder visible within the amniotic cavity (Figure 1), right hydronephrosis (Figure 2) and significant bowel dilatation suggestive of obstruction. The bowel diameter was 20×21 mm (normal 6–13 mm). Qualitatively, the bowel was well perfused, as determined by color power angiography of the loops in the amniotic cavity and the mesenteric blood flow; however, umbilical artery Doppler velocimetry showed reduced end-diastolic flow. The right kidney measured 60.5 mm (normal 34 ± 2 mm) in length (Figure 2) and the renal pelvis was markedly dilated (26×32 mm). The left kidney measured 40.7 cm. In the previous scans, the bladder had been identified in its normal intra-abdominal position.

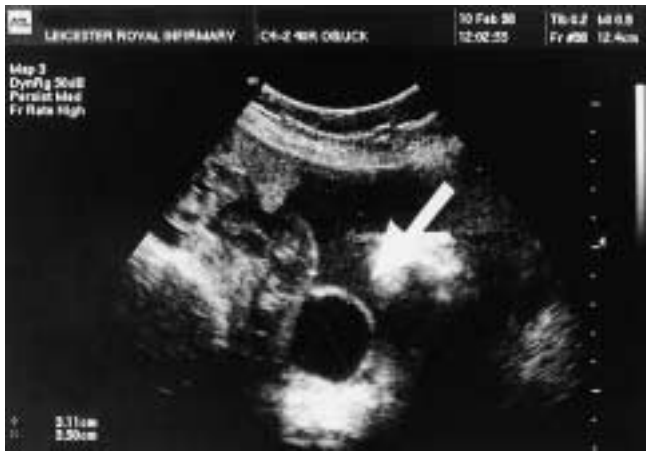

Figure 1 Eviscerated bladder (arrow) in the amniotic cavity with bowel superior to it

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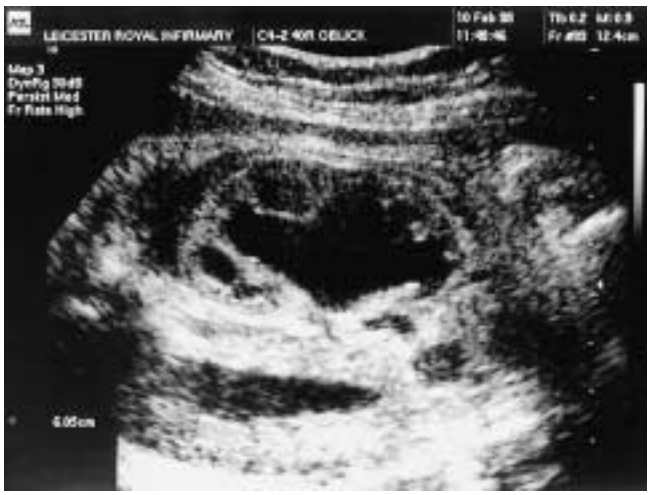

Figure 2 Longitudinal section of the right fetal kidney, showing hydronephrosis and dilated renal pelvis (kidney length 60.5 mm)

In view of the findings, the mother was given dexamethasone and the baby was delivered by Cesarean section 36 h later. The female infant weighed 1.99 kg. Neonatal examination prior to surgery confirmed the antenatal diagnosis and associated bladder evisceration. The hernia was successfully repaired by primary closure on the same day. The postoperative period was uneventful. Postnatal ultrasound scanning of the kidneys showed right hydronephrosis with marked right pelvic dilatation. The left kidney was slightly enlarged but its architecture was normal. A micturating cystourethrogram was normal and with no vesicoureteric reflux. The baby was discharged back to its local hospital 3 weeks after delivery and is being followed by pediatricians.

DISCUSSION

Gastroschisis is a paraumbilical abdominal wall defect, usually right sided, through which intra-abdominal contents, commonly the small and large bowel and stomach, eviscerate. Other structures that may eviscerate include the liver and spleen[3], the Fallopian tubes, ovaries, testicles and bladder[4]. Recognition of this latter group has, however, been mainly postnatal. The incidence of gastroschisis is reported to have risen from 0.67 per 10 000 births in 1987 to 1.35 per 10 000 births in 1991 in England and Wales[1]. It is more common in young mothers, especially those below the age of 20 years[1,2,5].

The etiology is uncertain but it has been suggested that it may be due primarily to intrauterine interruption of the omphalomesenteric artery[6]. An abnormal involution of the umbilical vein, resulting in a paraumbilical defect through which the small bowel prolapses at approximately 37 days of embryonic life[7], or alternatively an abnormality in the development of the somites responsible for the integrity of the anterior abdominal wall[8] have also been suggested as possible mechanisms. Cigarette smoking may cause an interruption in the omphalomesenteric arterial blood supply; hence the defect is more common in smokers[1]. Other factors implicated in being associated with an increased risk include use of cocaine, marijuana, amphetamines, alcohol[9,10] and the oral contraceptive pill[11].

Partial bladder evisceration was first reported by Diller and Travis[12], and Moore[8] reported full evisceration. In a review of 69 cases of gastroschisis by Novotny and colleagues[4], 26% had associated anomalies; bladder evisceration occurred in three cases (4.3%). Kirk and Wah[13] also reported a case in which, in addition to the gastroschisis, there was evisceration of the liver, spleen, bladder and both testes. In all these cases, however, the evisceration was diagnosed postnatally. The findings of bladder evisceration complicated by hydronephrosis, diagnosed prenatally using ultrasound in this case and that reported by Tannouri and co-workers[14] , reinforce the need for continual monitoring of the fetus after diagnosis.

Once a diagnosis of gastroschisis has been made, it is imperative to search meticulously for associated abnormalities, as some of these may significantly alter the prognosis and management of the baby. Associated malformations occur in 5–23% of cases of gastroschisis compared to 66% in cases of omphalocele[15,16]. Most are jejunoileal or colonic atresia. The mortality rate in gastroschisis is about 12% compared with 34% in omphalocele. Most of these occur in patients with jejunoileal atresia, bowel necrosis or motility dysfunction[5]. Long-standing exposure of the bowel to amniotic fluid may cause motility dysfunction due to inflammation and shortening of the intestinal loop[5]; therefore there is a need to search for ultrasound features that would help in predicting outcome.

The presence of other herniated structures, such as the bladder, in this case, may not necessarily alter the management, but the occurrence of complications secondary to such herniation may. In this case, significant hydronephrosis of rapid onset was the relevant determining factor for early delivery. Hydronephrosis that was worse on one side suggested that there might have been kinking or obstruction at the trigone due to the position of the bladder. Although there is currently no proven quantitative method of assessing bowel perfusion, our experience (unpublished) suggests that poor qualitative perfusion in association with dilatation is often followed by repeated bowel problems in the neonatal period. Delivering before the onset of reduced perfusion in the presence of bowel dilatation therefore improves the prognosis.

This case would suggest that, after the initial diagnosis of gastroschisis and exclusion of other abnormalities, continuing ultrasonographic surveillance of the fetus should be maintained, as other structures that might affect management and prognosis may eviscerate later. Failure to identify such complications may have significant medicolegal implications. We therefore suggest that such surveillance is as detailed as possible. Although bladder evisceration may not affect the prognosis, it may be complicated by hydronephrosis due to ureteric obstruction. We speculate that, if this is not relieved early, it may cause significant renal damage.

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