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The outcome of gastroschisis after a prenatal diagnosis or a diagnosis only at birth Recommendations for prenatal surveillance

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Abstract

Objectives: To establish in infants with gastroschisis whether outcome is different when comparing a prenatal diagnosis with a diagnosis only at birth with the intention to develop a prenatal surveillance protocol. Intestinal atresia established after birth and preterm versus term delivery were studied as risk factors.
Study design: All 24 fetuses and 9 infants diagnosed with gastroschisis and referred to our tertiary center between January 1991 and June 2003 were studied retrospectively.
Results: The infants of the prenatal subset delivered at our tertiary center and 18 survived. There were two pregnancy terminations, three intrauterine deaths at 19, 33 and 36 weeks respectively and one neonatal death. All nine infants in the postnatal subset survived. Eight were out born and one was delivered at our tertiary center. Prenatal bowel dilatation did not correlate with outcome. Between the prenatal and postnatal subset no significant difference in outcome of live-born infants was established. For four infants with intestinal atresia a significant difference was demonstrated for induction of preterm labour ($P < 0.05$), duration of parenteral nutrition ($P < 0.01$), number of additional surgical procedures ($P < 0.001$) and length of hospital stay ($P < 0.01$). The fifteen infants born prior to 37 weeks of gestation spent a significantly longer period in hospital compared to those delivered at term. When the cases with bowel atresia were excluded this difference was no longer present. Five of the 33 cases were diagnosed with associated anomalies which mainly involved the urinary tract.
Conclusion: Neonatal outcome of live born infants following a prenatal diagnosis of gastroschisis is not different from a diagnosis at birth. The presence of intestinal atresia is the most important prognostic factor for morbidity. The supplemental value of prenatal diagnosis to the outcome of infants with gastroschisis may be in the prevention of unnecessary intrauterine death and detection of intestinal complications. A proposed surveillance protocol for fetuses with gastroschisis focused on intrauterine signs of pending distress such as a dilated stomach, intra abdominal bowel dilatation with peristalsis, notches in the umbilical artery Doppler signal, development of polyhydramnios and an abnormal CTG registration may improve outcome.

Keywords: Gastroschisis; Intestinal atresia; Prenatal diagnosis; Fetal outcome; Fetal surveillance

1. Introduction

Gastroschisis is a congenital defect of the abdominal wall characterized by the evisceration of abdominal organs without a covering membranous sac.

The anomaly can easily be detected by prenatal ultrasound. The detection rate has been reported to be as

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high as 83%, with the majority being diagnosed before the third trimester [1].

Infants with gastroschisis have a high overall survival rate, but the rate of intra uterine fetal death and morbidity due to gastrointestinal complications is considerable [2]. Bowel dilatation [3] and polyhydramnios [4] have been suggested as prenatal predictors of adverse postnatal outcome, although for the size of the dilated bowel [5] this may be questioned. Preterm delivery and particularly pre-labour caesarean section may result in a reduction in the formation of a fibrous coating on the bowel as well as reduce complications such as atresia, stenosis, necrosis and perforations of the bowel [6]. However there is still controversy regarding the use of caesarean section and preterm instead of term delivery [7]. Several studies have come to the conclusion that term vaginal delivery will improve outcome [8,9]. The positive effect of prenatal diagnosis on the outcome of gastroschisis has not been established, although these studies were conducted 8–10 years ago [10–12].

Routine ultrasound to detect congenital anomalies was not part of recent policy in the Netherlands therefore some cases of gastroschisis remained undiagnosed in the antenatal period. This provided us with the opportunity to assess the value of prenatal diagnosis for fetuses with gastroschisis and to propose a surveillance protocol according to outcome parameters and data from the literature. We studied (i) the influence of prenatal diagnosis on the outcome of fetuses with gastroschisis compared with a group of infants where this diagnosis was only established at birth, (ii) the influence of bowel atresia on outcome of infants with gastroschisis and (iii) the outcome of infants with gastroschisis born before or after 37 weeks of gestation.

2. Material and methods

The University Medical Centre Rotterdam serves as the referral centre for both fetal anomaly scanning and paediatric surgery in the South West of the Netherlands. We performed a retrospective analysis of all infants from singleton pregnancies diagnosed with a gastroschisis either pre or postnatally between January 1991 and June 2003. Cases were identified from our ultrasound database and from the patient database of the Department of Paediatric surgery.

Following prenatal detection of a gastroschisis, regular ultrasound scans were performed during pregnancy to evaluate fetal growth, the presence of bowel dilatation and amniotic fluid volume. During the study period no structured protocol was applied to monitor the fetal condition. Fetal growth restriction (FGR) was defined as a fetal upper abdominal circumference $<$10th centile. Bowel dilatation was determined by the inner-to-inner bowel wall distance. Maximum bowel dilatation prior to delivery was stratified employing a cut-off level of 10 and 17 mm and correlated with outcome. Amniotic fluid volume was considered abnormal for measurements below the 5th and above 95th percentile [13]. Counseling by a paediatric surgeon was offered to all couples during the prenatal period. According to the hospital protocol all women with a prenatal diagnosis of gastroschisis were delivered in our centre by vaginal delivery unless obstetric reasons required otherwise. Prematurity was defined as a delivery prior to 37 weeks of gestation. Infants with a birth weight below 10th percentile, adjusted for sex [14], were defined as small for gestational age (SGA).

After delivery the presence or absence of a fibrous coat on the bowel (bowel peel) was documented. Management of the gastroschisis consisted of closure of the defect within hours following birth, by means of primary closure or Siliastic Silo depending on the size of the defect and ventilatory pressures during the procedure. Due to motility disorders of the bowel infants were given primarily parenteral nutrition (TPN) and enteral feeding was instituted as soon as stomach retentions diminished.

Adverse neonatal outcome was defined as neonatal death or complications resulting from the abnormality itself (intestinal atresia, necrotising enterocolitis), the subsequent surgical procedure (anatomic or functional short bowel syndrome, TPN associated cholestasis jaundice, venous line sepsis), or other significant morbidity not directly related to the gastroschisis.

Distinction was made between: (i) a diagnosis prenatally and a diagnosis only postnatally, (ii) presence and absence of intestinal atresia and (iii) delivery before and after 37 weeks of gestation. Charts were reviewed for maternal demographic data, fetal ultrasound surveillance data, fetal outcome, neonatal surgical and subsequent outcome data.

Statistical analysis of comparing groups was performed using the Mann–Whitney test or Fisher's exact test in case of continuous or categorical data, respectively. $P = 0.05$ (two-sided) was considered as the limit of statistical significance.

3. Results

We reviewed the data of 24 prenatally diagnosed and 9 postnatally diagnosed cases of gastroschisis. Three prenatal cases have been described previously [15].

In the prenatal subset, mean maternal age ($\pm$ S.D.) at diagnosis was 25.9 ± 6 years. Mean gestational age at diagnosis was 23.3 ± 6.6 weeks. In six out of 24 women (25%) the gastroschisis was detected after 24 weeks gestation. Two pregnancies were terminated before 24 weeks gestation. Three women were diagnosed with FGR. Five pregnancies were complicated by oligohydramnios and two by polyhydramnios. Associated fetal anomalies were detected in 4/22 (17%) pregnancies, unilateral hydronephrosis in three cases and bilateral talipes equinovarus in one. Three fetuses (12.5%) died in utero at 19, 33 and 36 weeks of gestation, respectively. One intra uterine death represented a combination of gastroschisis, bilateral talipes