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Research paper

Gastroschisis: methods and early outcomes of surgical treatment

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Abstract

Introduction: Gastroschisis is a congenital anomaly in which loops of intestine are displaced outside the abdominal cavity through a defect in the abdominal wall. The incidence is steadily increasing. It is particularly attributed to young maternal age. The defect requires urgent surgical intervention after birth.

Aim: The article aims to present the methods and early outcomes of surgical treatment of new-borns with gastroschisis in the context of pre- and postnatal examinations, and attempts to identify risk factors for the occurrence of the defect.

Material and methods: The study used data derived from medical records of 18 patients of the Regional Specialist Children's Hospital in Olsztyn. Prenatal, peri- and postoperative outcomes were analysed. The new-borns were classified with simple and complex gastroschisis.

Results and discussion: The defect was diagnosed prenatally in 15 new-borns. In 16 cases, gestation was terminated by caesarean section. The mean maternal age at birth was 23 years; 14 woman were pregnant for the first time. All new-borns were operated on on day 1 of life. In 14 patients, primary closure of the abdominal wall was performed. The mean duration of mechanical ventilation was 13.31 days, of total parenteral nutrition – 29 day, to the first administration of enteral nutrition – 11.88 days and the mean length of hospital stay was 39.75 days, in the group of infants with simple gastroschisis.

Conclusions: Gastroschisis is a severe anomaly causing an immediate threat to life. The optimal treatment procedure is the primary closure of the abdominal wall. The use of silastic sac enables the staged closure. Risk factors include young maternal age and primiparity.

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1. INTRODUCTION

Gastroschisis is a congenital anomaly in which loops of intestine are displaced outside the abdominal cavity through a defect in the abdominal wall, usually located on the right side of the attachment of the normally developed umbilical cord.[1] The displaced intestines are not covered with foetal membranes and are exposed to the effects of the amniotic fluid (Figure 1).

The incidence of gastroschisis is estimated at 1 to 4 per 10 000 live births, irrespective of gender, and the number of cases is steadily increasing.[1] The increasing defect incidence is particularly attributed to environmental risk factors, of which young maternal age is considered the most relevant.[2,3] In addition to maternal age, the following are also considered: primiparity, use of medicines and of cocaine, alcohol consumption, and smoking cigarettes by pregnant women.[2] Congenital gastroschisis occurs at an early stage of embryogenesis, usually between week 4 and 10 of intrauterine life.[4] Etiopathogenesis is not clear. The causes are attributed to the premature regression of the omphalomesenteric artery or the right umbilical vein and to disturbances at the stage of lateral mesenchymal fold migration.[1]

A characteristic feature of gastroschisis is inflammation of the intestinal wall, which increases its volume. The extent and severity of the inflammatory process result from impaired blood supply to the intestines, mechanical injury, and exposure of the intestinal wall to the substances contained in the amniotic fluid, such as urine and pro-inflammatory cytokines.[5]

Most cases of gastroschisis are diagnosed prenatally during a medical ultrasound examination after week 12 of foetal life.[5–7] In most facilities, the diagnosis is made in the second trimester.[5,6,8] A prenatal diagnosis of gastroschisis is an indication for caesarean section.[9] Regardless of the time of diagnosis, the defect requires urgent surgical intervention after birth.

The surgery is performed using one of two surgical techniques. The choice is determined by the extent of the gastroschisis, local bowel condition, the new-born's abdominal cavity volume and the surgeon's preference. The optimal surgical method is a one-stage repair of the abdominal wall.[1,10] When all other options for performance of the above surgery have been excluded, multi-stage treatment using a silastic sac is applied to avoid, for example, increased intra-abdominal pressure syndrome. The staged method was initiated by Schuster and with variations, is still applied to date.[11] In the treatment of giant gastroschisis, the vacuum-assisted closure (VAC) technique is applied.[12] This enables the gentle approximation of wound edges, activates molecular pathways of angiogenesis and cell division, and it allows oedema to be reduced due to the ability of VAC to remove large amounts of fluid.[13]

New-borns with congenital gastroschisis require specialist postoperative care, including mechanical ventilation (MV), total parenteral nutrition (TPN), and the treatment of possible complications. The survival rate of new-borns

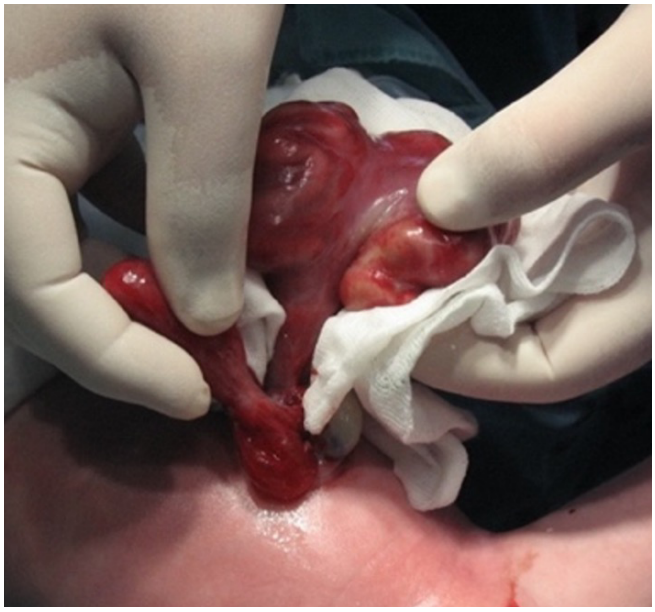

Figure 1. Intestinal gastroschisis. Own archive'.

born with gastroschisis has been steadily increasing,[7] and the prognosis is good.[14] There are two forms of gastroschisis: isolated and complex (with accompanying intestinal obstruction and underdevelopment). Cases of complex gastroschisis are less common and are characterised by a poorer prognosis of survival and a risk of development of complications, e.g. short bowel syndrome.[15]

2. AIM

This article aims to present the methods and early outcomes of surgical treatment of new-borns with gastroschisis in the context of pre- and postnatal examinations, and attempts to identify risk factors for the occurrence of the defect.

3. MATERIAL AND METHODS

The study used data derived from medical records of patients of the Clinical Ward of Paediatric Surgery and Urology of the Regional Specialist Children's Hospital in Olsztyn. In the years 2011–2020, 19 new-borns (8 girls and 11 boys) were treated for gastroschisis in the ward. However, due to incomplete hospital records of one of the patients, the medical histories of only 18 new-borns were subjected to analysis.

The following were analysed: time of diagnosing the defect, delivery method, infant's gender, foetal age at birth and the number of hours between birth and surgery. The choice of surgical treatment method and intestinal patency were also analysed. Those new-borns in whom no signs of obstruction were found were classified with simple gastroschisis (SG), while those whose bowel condition demonstrated signs of obstruction were classified with complex

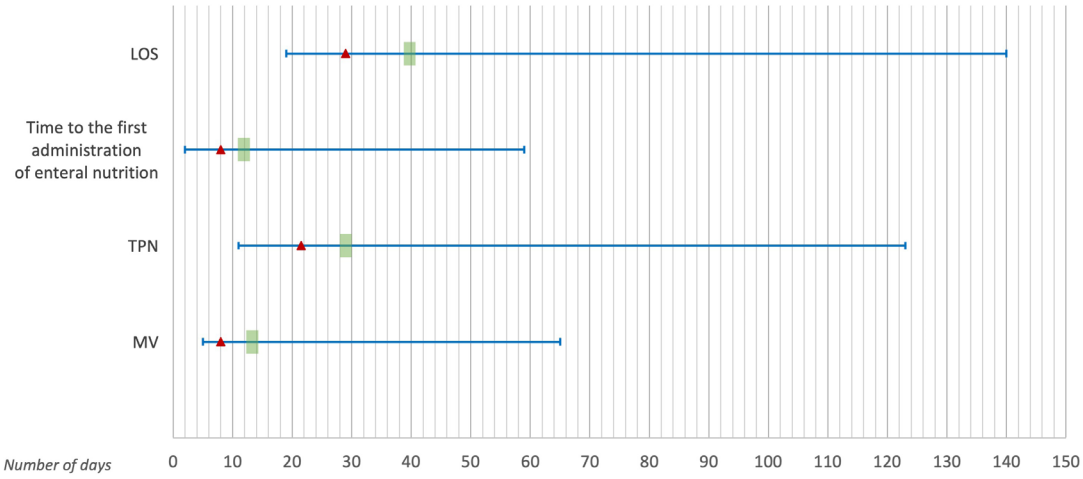

Figure 2. Postoperative outcomes.

gastroschisis (CG). The following were classified into the data concerning postoperative outcomes of new-borns: number of postoperative MVs, number of TPN days, time to the first administration of enteral nutrition and length of hospital stay (LOS). The above data analysis was limited to new-borns with SG, as only 2 cases with CG were noted. The outcomes of new-borns with CG are not included in the diagram. The attempt to identify risk factors for the occurrence of the defect was limited to an analysis of the maternal age at birth and the sequence of pregnancy.

4. RESULTS

In the years 2011–2020, in the Clinical Ward of Paediatric Surgery and Urology of the Regional Specialist Children's Hospital in Olsztyn, 19 new-borns (8 girls and 11 boys) were surgically treated for gastroschisis, with an average of 2 surgeries per year. The medical records of 18 infants were analysed.

The defect was diagnosed prenatally in 15 (83.3%) new-borns. In 16 cases (88.9%), gestation was terminated by caesarean section (C/S), and in 2 cases, (11.1%) by normal spontaneous vaginal delivery (NSVD). Ten (58.8%) were born before week 37 of gestation, and 7 (41.2%) after week 37 of gestation; in 1 (5.6%) infant, the foetal age at birth was unknown. The mean maternal age at birth was 23 years; 10 (55.6%) pregnant women were less than 25 years, and 8 (44.4%) were 25 years or more; 14 (77.8%) were pregnant for the first time, 1 (5.6%) for the second time, and 3 (16.7%) for the third time. One of the mothers, who was pregnant for the third time, gave birth for the first time. In the rest of the cases, the number of pregnancies corresponded to the number of births. The mean birth weight was 2653.53 g; 6 (35.3%) patients weighed less than 2500 g, 11 (64.7%) weighed 2500 g or more; 1 new-born was not weighed at birth. All new-borns were operated on on day 1 of life, on average at 5 h after birth; in 5 (29.4%) cases, the procedure was performed 3 h after birth, in 12 (70.6%) after 3 h. In 1 (5.6%) case, the time from birth to surgery could not be determined.

In 14 (77.8%) patients, primary closure of the abdominal wall was performed, while staged treatment was applied in 4 (22.2%) patients. In 2 (11.1%) patients, features of intestinal obstruction were found during the procedure. One (5.6%) new-born required the removal of sections of the small intestine and the externalisation of double-barrel stoma, and 1 (5.6%) required the removal and end-to-end anastomosis of intestinal sections and the externalisation of the appendicocecostomy. All the infants survived.

In the postoperative period, all the infants required MV, on average for 13.31 days in the group of new-borns with SG. The duration of MV for the 2 patients with CG was 6 and 32 days. All patients received parenteral nutrition; the mean duration of TPN was 29 days, and the mean time to the first administration of enteral nutrition was 11.88 days in the group of infants with SG. The duration of TPN in the 2 infants with CG was 55 and 94 days, while the time to the first administration of enteral nutrition was 7 and 8 days, respectively. The mean LOS was 39.75 days for new-borns with SG. The LOS for infants with CG was 55 and 96 days. Figure 2 shows postoperative outcomes, including the mean (green rectangle), median (red triangle) and the minimum and maximum number of days of MV, TPN, the number of days to the first administration of enteral nutrition and LOS in the group of infants with SG.

5. DISCUSSION

In the years 2011-2020, in the Regional Specialist Children's Hospital in Olsztyn, 19 new-borns were treated surgically for gastroschisis, with an average of 2 surgeries per year. An analysis of the collected data shows a relative plateau in the incidence of gastroschisis, although numerous publications indicate a global increase in the incidence of gastroschisis over the past decades.[3,16,17] However, regarding the incidence of gastroschisis, it should be noted that the small number of recorded cases in the past might have been due to the lack of uniform nomenclature for abdominal wall defects and to difficulties in differential diagnosis related to the omphalocele.[3]

With the development of new imaging techniques and their increasing availability, many anatomical foetal defects can be diagnosed prenatally. The current study found that, for gastroschisis, the defect was diagnosed in most new-borns (83.3%) before birth. Recent reports confirm that prenatal diagnoses outnumber postnatal diagnoses.[6,7,15,18,19]

Prenatal diagnosis of gastroschisis is considered in the literature in the context of the benefits that early detection of the defect could bring and the possible implementation of therapeutic steps during the course of pregnancy. Researchers' views on the improvement of the prognosis and postnatal outcomes of new-borns are rather sceptical.[6,8] However, prenatal diagnosis of the defect should prompt the referral of pregnant women to reference centres in which the appropriate care of paediatric surgeons and neonatologists, as well as genetic counselling, would be provided.[14] As regards prenatal therapeutic interventions, no beneficial effects were observed of steroid therapy on the postoperative outcomes of new-borns with gastroschisis.[20] Moreover, Luton et al., despite having noted the contribution of inflammation to the pathogenesis of gastroschisis, emphasise that amniotic fluid replacement offers no benefits in terms of clinical condition or the development of complications.[21]

An unresolved but constantly debated issue is the optimum route and timing of delivery. Most of the new-borns (88.9%) from the analysed study group were born by C/S. Similar observations were also made by researchers from Polish and other centres.[1,8,15,17,22] It is believed that C/S can reduce the probability of damage to the intestines and mesentery, or the occurrence of short bowel syndrome.[15,23] Moreover, this delivery method can ensure more effective cooperation between specialists in various fields in the area of perinatal care.[1] However, most studies conducted to date have not confirmed any significant benefit of C/S.[22,24] No correlation was demonstrated between the route of delivery and the mortality of new-borns with gastroschisis.[22,25] It is therefore recommended that the choice of delivery method be left to the obstetrician and the mother, and the C/S procedure be performed for obstetric indications as recommended by gynaecological associations.[5]

To date, no consensus has been reached on the timing of birth either. The group of patients in the current study was dominated by the group of new-borns born before week 37 of gestation (58.8%). Several observations have been made by other researchers.[17,26] There have been hypotheses in the literature that an earlier delivery may reduce intestinal damage and improve postnatal outcomes.[27] These authors, however, are inclined to conclude that delivery before week 37 of gestation offers no significant benefits in terms of the prognosis.[19,28] On the other hand, Boutors et al. demonstrated a decrease in the LOS and the number of MV and TPN days in infants with gastroschisis with an increase in the gestational age and birth weight of the foetus at birth.[24] This may be related to greater foetal maturity and a decrease in the number of complications resulting from preterm birth. This is, therefore, an important counterargument to planning preterm delivery. In the authors' opinion, it is necessary to respect the existing recommendations in which premature delivery in the case of gastroschisis is not recommended.[5,19,24]

In the current analysis, the number of mothers who gave birth to infants with gastroschisis at the age of less than 25 years (55.6%) was greater than the number of mothers who gave birth at the age of 25 years and more (44.4%). The results obtained, in which the cut-off criterion for a maternal age of 25 years is adopted, are consistent with the results of studies by other researchers.[26,29] Anderson et al. clearly indicate that the probability of defect occurrence decreases with maternal age.[17] Some authors observed a statistically significant frequency for the development of the abdominal wall defect in new-borns of mothers under 20 years of age.[17,25] In the current study, there were only 4 (22.2%) women in this group. There is also evidence that primiparity is a statistically significant risk factor for gastroschisis.[29] The highest risk of giving birth to an infant with gastroschisis was demonstrated for primiparas. This is confirmed by the current study, i.e. 14 (77.8%) women were pregnant for the first time.

The issue of differences in the maternal cut-off age, at which the probability of giving birth to an infant with gastroschisis is particularly high, may also arise from an increase in the mean age of primiparas. Data published by the European Statistical Office (Eurostat) indicate that in 2018, women gave birth to their first child at an average age of 30.8 years, i.e. approximately a year later than in 2007. It is, therefore, possible that the continuation of this upward trend may lead to an increase in the probability of the occurrence of gastroschisis even in infants of mothers giving birth after 25 years of age.

In the Regional Specialist Children's Hospital in Olsztyn, primary closure of the abdominal wall was performed more frequently (77.8%). One-stage reduction of the intestines and multi-layer reconstructive operation of the abdominal wall is the predominant method in many facilities.[10,18,19,30] Banyard et al. demonstrated that for patients in which the primary closure of the abdominal wall had been performed, the duration of MV, TPN and LOS were shorter than for the patients who had been treated using silastic sac. They suggest that the LOS-affecting parameter is primarily the number of days in which the intestines are maintained in a sac.[10] On the other hand, Pastor et al. observed a trend towards a reduction in the number of days of MV and the intra-abdominal pressure where the intestines had been placed in a silastic sac. Despite the small size of the study group, they concluded that multi-stage treatment using a Silastic pouch can be as effective and safe as the primary closure of the abdominal wall.[18]

The development of prenatal diagnosis and an improvement in peri- and postoperative care, primarily in high-income countries, have resulted in a predominance of live births among new-borns with gastroschisis, and the mortality rate remaining at a low level (approximately 4%).[6,7,17,31] In the study group of new-borns operated on at the Regional Specialist Children's Hospital, all patients with the described gastroschisis survived.

Attempts are still being made to identify factors that are likely to have an effect on the survivability of infants with gastroschisis, as the mortality rate for these patients is higher than for normal pregnancies.[14] However, certain facilities have seen a decrease in the number of deaths over the last 20 years.[1,7] The last death of a new-born with polyhydramnios and gastroschisis in the Regional Specialist Children's Hospital was in 2004.

Some studies indicate that the factors which increased the risk of death independently of each other included the time of delivery before week 37 of gestation and too low birth weight in relation to the foetal age, which is inherently associated with preterm birth and its complications.[25] However, Anderson et al. noted no difference in the mortality of new-borns weighing less than 2500 g compared to intants with a bodyweight appropriate for the foetal age, yet they observed increased mortality among patients born before week 37 of gestation.[17] Moreover, researchers studying cases of gastroschisis conclude that CG involves a greater risk of death than SG.[8,15] These research findings indicate that further multi-centre studies are necessary to carry out a statistical analysis and verify the current state of knowledge of gastroschisis.

6. CONCLUSIONS

(1) Gastroschisis is a severe developmental anomaly causing an immediate threat to life.
(2) Prenatal diagnosis and monitoring of the course of pregnancy and birth offers the possibility of planning the time, location and method of intervention in the first hours of life. It determines the treatment outcome, which, in turn, depends on the tactics adopted and the facility's experience, too.
(3) The optimal procedure in the treatment of gastroschisis is the primary closure of the abdominal wall.
(4) The use of silastic sac enables the staged closure of gastroschisis.
(5) Risk factors for the occurrence of gastroschisis include young maternal age and primiparity.

Conflict of interest
None declared.

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Ethics
It was a retrospective study hence no institutional ethics committee's approval was required as well as no written informed consent was obtained.

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