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Laparoschisis : About A Case Report And Literature Review

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Abstract: *Laparoschisis is a congenital anomaly characterized by the protrusion of abdominal organs, predominantly the intestines, through a defect in the abdominal wall, typically observed on the right side. It is categorized into simple and complex types based on the extent of the defect and involvement of other organs. The estimated prevalence of laparoschisis is approximately 1 in 10,000 births, with variations noted in different regions. Interestingly, certain areas, like New York and Georgia exhibit an increasing trend, particularly among younger mothers. In Morocco, a study carried out at the "Les Orangers" hospital in Rabat in 2020 showing that the laparoschisis represent 5.1% of musculoskeletal malformations in newborns (equivalent to 1.7per 10,000 total births). Prenatal ultrasound is the primary diagnostic tool, enabling early detection of the abdominal wall defect and protruding organs. Magnetic resonance imaging (MRI) can provide additional information for diagnosis. Treatment options for laparoschisis include primary repair, performed shortly after birth, and delayed closure, which is postponed for a certain period after birth, depending on case-specific factors such as defect size, condition of protruding organs, and the presence of complications such as stenosis and atresia. Prognosis is influenced by the gravity of the malformation and associated conditions, necessitating close obstetrical and neonatal monitoring with tailored interventions for optimal outcomes. To illustrate the practical aspects of laparoschisis management, we present a case study involving a live-born infant diagnosed with laparoschisis at 34 weeks' gestation using prenatal ultrasound. A comprehensive one-stage surgical repair was performed on the newborn. This case highlights the importance of early antenatal diagnosis through ultrasound, potential complications, and immediate and secondary management strategies for optimal outcomes in affected infants.*

Introduction

Laparoschisis is a congenital malformation distinguished by the protrusion of digestif organs through a defect, often in the right side of the abdominal wall. Its prevalence varies across geographical regions and populations. While remaining relatively uncommon. its incidence has shown an upward trend in recent years, ranging from approximately 2 to 4 cases per 10,000 live births in North America and Europe. The factors that increase the risk of this malformation include being under the age of 20, smoking, as well as the use of drugs and alcohol[1].

Although the physiopathology is not well known, however, several embryological theories have been suggested, the most recent of which is proposing that thrombosis in the vicinity of the umbilical ring, induced by the natural involution of the umbilical vein, could potentially weaken the structure of the umbilical ring, thereby predisposing it to herniation [2].

Imaging tests such as ultrasound and MRI enable us to make an accurate prenatal diagnosis, assess the severity of the condition, and detect complications such as atresia and necrosis. A number of cases in the literature report on the usefulness of antenatal dignotics in intrauterine surgery, and emphasize its value.

When considering the diagnosis, which is a more effective diagnostic tool for detecting laparoschisis: MRI or ultrasound?

When faced with the diagnosis, what factors should be considered when deciding between vaginal delivery and a Cesarian section?

In our article, we report the case of a live-born infant born with laparoschisis at 35 weeks' ammennorhea who was diagnosed by a prenatal ultrasound at 34 weeks' ammennorhea. In the following days, he underwent a complete one-stage surgical repair.

In our case, we highlight the importance of antenatal diagnosis, the complications that can accompany the pathology, as well as immediate and secondary management.

Case presentation

Our case is about a 27-year-old woman, with no significant pathological history, Gestation 3 Para 2, with a current unattended pregnancy, estimated at 35 weeks of amennorhea according to the date of the last accurate menstrual period, from a non-consanguineous marriage with a 27-year-old man, also with no significant pathological history.

Her first two deliveries were vaginal, giving birth to two girls, currently aged 6 and 3, with good psychomotor development and no congenital malformations.

The parturient was initially seen at 34 weeks of amenorrhea and an obstetrical ultrasound was performed showing a monotonic progressive pregnancy with positive cardiac activity. The amniotic fluid was normal in quantity and the cord contained 3 vessels. Morphological ultrasound of the anterior abdominal wall showed an 8.8 mm right parabolic defect with herniation of the entire digestive tract into the amniotic fluid without a sac, with a normal echogenicity, in addition to a 24 mm dilated digestive tract with no parietal thickening and no other intra-abdominal digestive dilatation. The rest of the ultrasound was unremarkable [figure (1)].

4 days later, the patient was admitted to the maternity department with an emergency premature delivery. On general examination, her vital signs were found to be normal. On obstetrical examination, the cervix was dilated to 7 cm, with a fixed cephalic presentation, a ruptured water sac with clear fluid, and a clinically normal pelvis.

Fetal heart rate was regular with142 beats per minute.

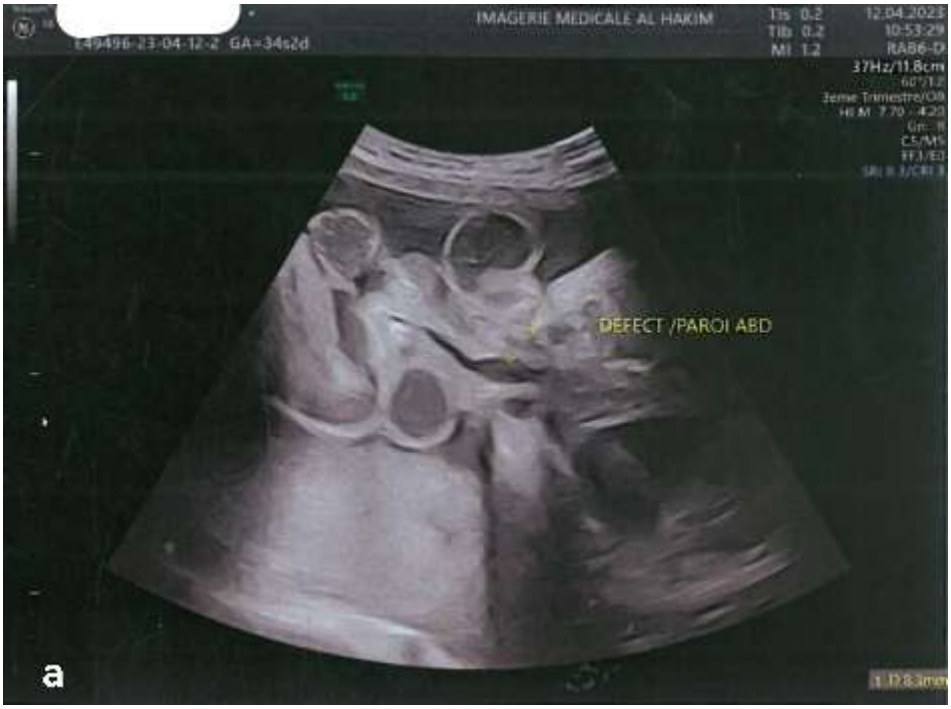
The vaginal delivery was eutocique, without episiotomy and without instrumental extraction, giving birth to a premature female newborn, with a birth weight of 2700g. Her APGAR score was 10/10 at the 5th minute. She showed a right-sided laparoschisis with a normally inserted umbilical cord. Intestinal tract did not reveal any atresia. There were no other abnormalities in the newborn [figure(2)].

Investigations were carried out on the newborn to look for complications of other malformations, like a full blood count, coagulation screening, as well as renal, liver, and bone profiles were conducted, all of which revealed normal results. An abdominal and kidney ultrasound scan was also conducted, which showed normal results.

Once the newborn's condition had stabilized, He was brought to the surgical room for medical intervention.The procedure entailed reducing the bowel and repairing the defect. Conducted under general anesthesia with a urinary catheter in place, the exploration revealed exteriorized and viable bowels. The defect was subsequently enlarged through a median incision.. An appendectomy was performed and was referred for pathological study.

The bowels were reintegrated in a complete common mesentery with verification of the intrabdominal pressure. A Delbet's blade was placed with skin closure and placement of a compressive bandage.

Short- and long-term respiratory, urinary and digestive outcomes were favorable. No complications were reported [figure (3)].



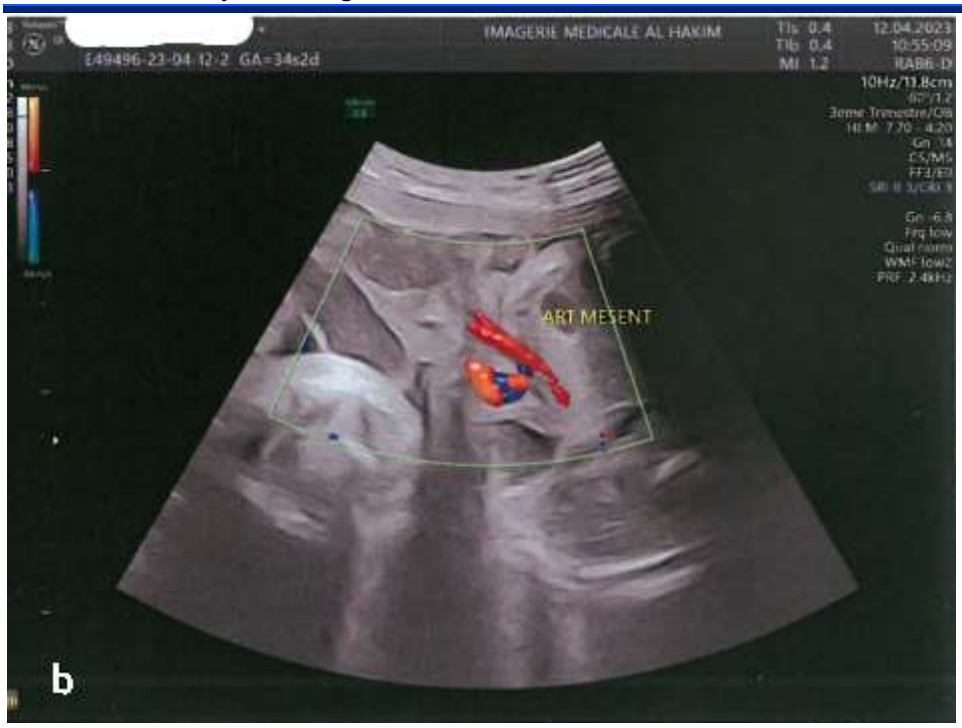


Figure [1] : Obstetric ultrasound at 34 weeks of amenorrhea **(a) :** showing at the level of the anterior abdominal wall of a right para-umbilical defect of 8,5 mm with exteriorization of almost all the digestive loops in the amniotic fluid without bag **(b)** : echo-doppler visualizes the mesenteric artery through the defect.

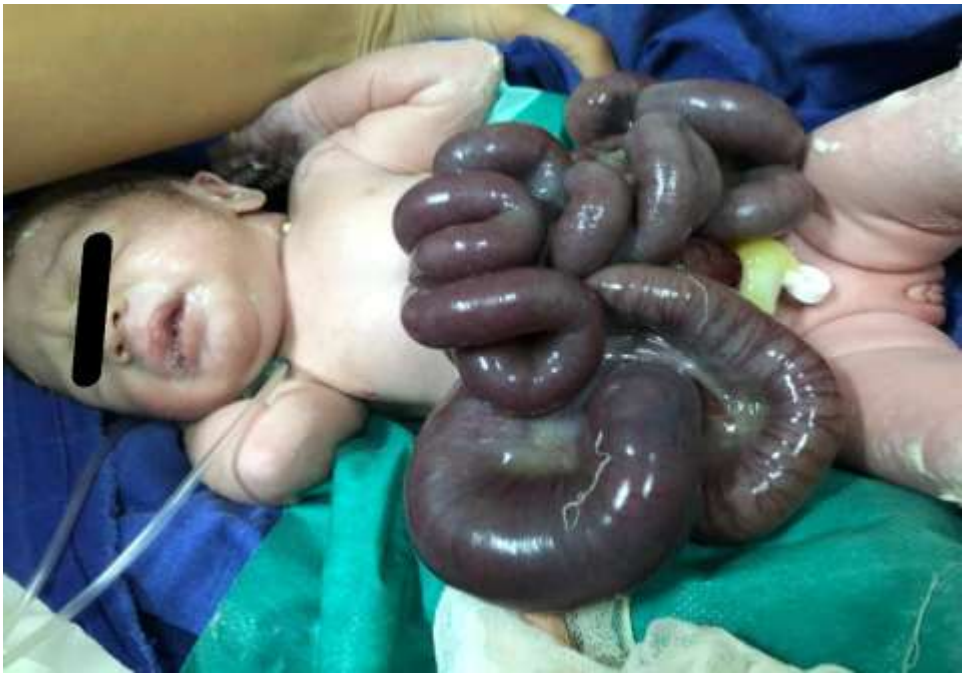


Figure [2]: the newborn immediately postnatal with exteriorization of almost all of the small intestine and colon through a right para-umbilical defect.

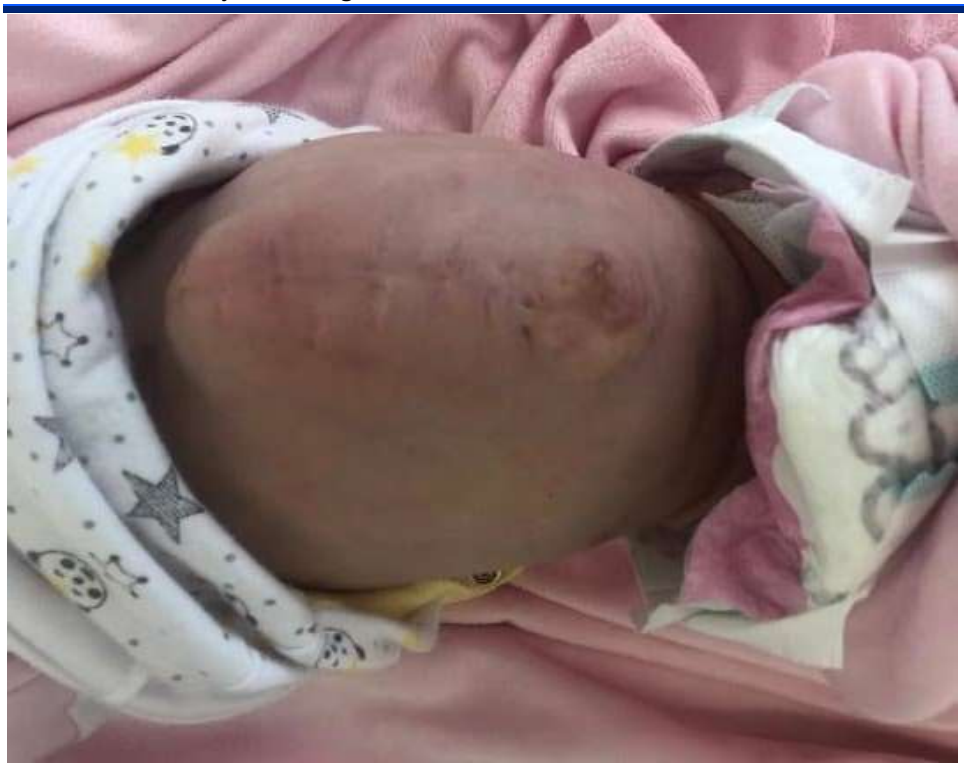


Figure [3]: the newborn at 45 days after surgical repair with good wound healing .

Discussion

Laparoschisis is a congenital malformation that involves the protrusion of the internal organs, typically through a defect in the right side of the abdominal wall, and without a protective membrane covering. Furthermore, a research done by Abdullah and Al. has demonstrated that around 10% of infants with laparoschisis experience intestinal stenosis or atresia[2], which is often caused by a lack of blood flow due to either volvulus or compression of the vascular pedicle by a narrow abdominal ring.

The exact cause of laparoschisis remains not fully understood. However, various embryological theories have been suggested, including abnormal right umbilical vein involution, failure of embryonic mesenchyme differentiation, omphalomesenteric artery interruption, amniotic membrane rupture, omphalomesenteric artery interruption, and amniotic rupture in the pars flaccida region of the umbilical cord[3]. More recently, a vascular-thrombotic model has been proposed, which suggests thrombosis near the umbilical ring caused by the normal involution of the umbilical vein may weaken the umbilical ring, creating a susceptibility to herniation. [4]

The incidence of laparoschisis has been on the rise in the United States during the last thirty years. Nonetheless, it's important to note that this trend is not consistent across all regions, as some areas have reported low and stable rates of laparoschisis. [1]. Laparoschisis tends to affect younger mothers disproportionately, with the highest occurrence seen among mothers who are under the age of 20. Factors such as maternal smoking, drug use, and alcohol consumption during the early stages of pregnancy, along with the father's younger age, have also been associated with laparoschisis[5]. In our case, none of these risk factors were found.

Molik and colleagues have classified laparoschisis into two categories: simple and complex types[6]. In simple laparoschisis, the intestines are in a healthy state with no complications, while congenital complications are seen in complex laparoschisis. Closed or closing laparoschisis is a subset of complex laparoschisis, where the abdominal wall defect begins to close around the protruding bowel, leading to a range of intestinal complications. According to a recent meta-analysis, infants diagnosed with complex Laparoschisis, which accounts for 17% of cases, exhibit significantly elevated mortality rates (16.67%) in contrast to infants with simple Laparoschisis (2.18%) [7].

Infants with complex Laparoschisis experience higher rates of morbidities, including delayed initiation of enteral feeds, prolonged time to achieve full enteral feedings, and increased reliance on parenteral nutrition (PN). They also face a big risk of complications such as necrotizing enterocolitis (NEC), short bowel syndrome (SBS) and sepsis.

These infants typically experience extended hospital stays and are more prone to being discharged while still requiring enteral tube feedings and parenteral nutrition (PN). Furthermore, left-sided laparoschisis, which occurs in the left paraumbilical region, is an

uncommon condition that is more frequently found in females. It is also linked to a higher occurrence of extraintestinal anomalies when compared to right-sided lesions[8]. In our case, the newborn had indeed a simple laparoschisis.

Laparoschisis is typically diagnosed using a prenatal ultrasound around 12-20 weeks into the pregnancy. The evaluation of laparoschisis involves assessing several key factors, including[9]:

- No covering membrane or sac.
- Location of the umbilical cord insertion to the defect, which is typically paraumbilical and predominantly right sided.
- Identifying any eviscerated organs.
- Examining the eviscerated bowel, including signs of dilation and/or thickening.
- Detecting any associated malformations that may be present alongside laparoschisis.

Although Laparoschisis is readily identifiable in prenatal ultrasonography due to its distinct characteristic of bowel loops freely suspended within the amniotic cavity without any protective membrane. Nevertheless, achieving a comprehensive morphological analysis requires the use of MRI, as the examination of intra-abdominal anatomy has been historically underemphasized.The addition of MRI to the diagnostic approach provides detailed information regarding the volume of the defect, its contents, and the presence of any complications, which significantly contributes to the postnatal management of Laparoschisis cases. The fine details obtained from MRI enhance the understanding of the condition, allowing for more precise decision-making and implementation of appropriate interventions. Fetal MRI demonstrates significantly higher accuracy compared to ultrasound (US) in confirming and precisely delineating the degree of small bowel obstruction. Additionally, MRI proves superior in distinguishing complete obstruction from luminal stenosis[10].

Elevated levels of alpha-fetoprotein (AFP) in both amniotic fluid and maternal serum, along with increased amniotic fluid acetylcholinesterase (AChE), have been linked to laparoschisis. In a study that compared 23 pregnancies affected by laparoschisis and 17 pregnancies with omphalocele, it was observed that during the second trimester, the serum AFP levels in the laparoschisis group were approximately 9.42 times higher than normal, and all pregnancies with laparoschisis exhibited increased levels of amniotic fluid AFP.[11].

In our case, the diagnosis was quickly made with a morphological ultrasound performed at 34 weeks of ammenorrhea, showing an 8.8 mm right parabombilical defect with herniation of all the digestive tracts into the amniotic fluid without a sac, with normal echogenicity, plus a 24 mm dilated digestive tract without parietal thickening or other intra-abdominal digestive dilatation. The rest of the ultrasonography was unremarkable. MRI and AFP assays were not performed due to lack of funding.

Monitoring fetal growth is crucial for infants diagnosed with Laparoschisis, as up to 60% of cases may exhibit growth restriction.

The occurrence of chromosomal anomalies in cases of isolated laparoschisis is in line with the baseline risk seen in the general population. However, when there are additional structural abnormalities beyond the intestines, the likelihood of chromosomal anomalies rises. In such cases, amniocentesis may be recommended to assist parents in making informed decisions and planning for the care of the newborn. [12]. Following Laparoschisis diagnosis, ultrasound evaluations should be conducted at 3 to 4-week intervals from 24 weeks gestation to track fetal growth and amniotic fluid volume[12]. Oligohydramnios, which is linked to fetal growth restriction, increases the risk of cord compression. Conversely, polyhydramnios may suggest the presence of bowel atresia. Importantly, the occurrence of growth restriction in fetuses with abdominal wall defects can serve as a predictive factor for worse neonatal outcomes.[13].

In our case, the diagnosis was made late and pathological follow-up was not performed. Ultrasound was not in favor of IUGR, and amniotic fluid was of normal quantity.

In pregnancies affected by laparoschisis, the occurrence of intrauterine fetal death (IUFD) is around 5%, a notably elevated figure compared to the rate observed in pregnancies without complications. This elevated risk of stillbirth may be attributed to various factors, including umbilical cord compression resulting from acute dilatation of the bowel outside the abdominal cavity, oligohydramnios, inflammation mediated by cytokines, volvulus (twisting of the bowel), and vascular compromise.[14].

Roughly 30 to 40% of cases affected by laparoschisis result in spontaneous preterm labor and delivery. This heightened incidence of preterm labor in these cases can be linked to high levels of pro-inflammatory cytokines, specifically interleukin-6 and interleukin-8, which are detected in the amniotic fluid[15]. Remarkably, there is an observed association between spontaneous preterm labor and more extensive damage to the bowel loops, bowel occlusion, and the presence of stained amniotic fluid. This correlation may arise from repetitive fetal vomiting of gastrointestinal contents into the amniotic fluid, resulting in heightened inflammation.[16].

In our case, delivery was indeed premature at 35 weeks' amenorrhea, but no serious intestinal lesions were detected and the amniotic fluid was clear.

Vaginal delivery is generally not contraindicated for infants with Laparoschisis based on the condition alone. The timing of delivery is determined by factors such as gestational age, ultrasound results (fetal growth profile and bowel appearance), and results from fetal testing. A multidisciplinary team, involving specialists focused on maternal and fetal care, neonatologists, and pediatric surgeons, collaboration is essential when determining a delivery plan, and it's crucial to consider patient-specific factors. Survey data on practice patterns indicate that approximately 40% of maternal-fetal medicine specialists opt for delivery at 37 weeks, while 30% prefer delivery at 39 weeks if the condition is stable[12]. The remaining specialists may choose to deliver before 37 weeks. In general, many specialists allow for a trial of vaginal delivery, except in cases where there is significant liver herniation, which may warrant a cesarean section. Some specialists also consider the lack of bowel dilation more than 25 mm after 37 weeks gestation as a factor in determining the need for continued gestation.[12]

The selection of the delivery mode did not exhibit a significant correlation with overall mortality, necrotizing enterocolitis (NEC), secondary repair, sepsis, short gut syndrome, time until full enteral feeding, or length of hospital stay. Nevertheless, a particular study revealed that a cesarean section was recognized as an independent risk factor for the development of respiratory distress.[16]. Therefore, it is generally not recommended to plan for a c-section in the absence of typical obstetric indications.

In our case, the vaginal delivery was eutocic and without complications. No indication for Caesarean section was found.

Neonatal management begins in the labor room. Special attention is given to loss of fluid through the uncovered bowel, which are significantly higher compared to healthy newborns. However, excessive fluid resuscitation should be avoided to prevent complications. The exposed bowel is protected using a bowel bag, and the blood flow and perfusion are assessed[15]. An orogastric tube is introduced for the purpose of stomach decompression, and intravenous access is established for antibiotics and fluids. Adjustments are made to the fluid administration to compensate for the fluid losses. Lastly, the airway is evaluated and maintained.

The primary objective of laparoschisis repair is to reposition the exposed bowel and organs into the abdomen while minimizing any potential injury or increased pressure. Two primary treatment options exist: primary repair and delayed closure involving a temporary silo and gradual reduction of bowel contents. Primary closure may involve operative suturing or closing the abdominal wall without sutures after repositioning the bowel. Interestingly, a randomized trial noted no significant differences in outcomes between primary closure and delayed closure methods.[14].

In our case, a primary repair was performed. The bowels were reintegrated the same day in a single stage and the defect was repaired. No further surgery was performed.

Maintaining the integrity of the umbilicus during closure results in favorable cosmetic outcomes. A study that compared sutureless closure to sutured repair indicated longer recovery times and hospital stays, increased antibiotic usage, fewer cases of infection, and reduced time spent on a ventilator. However, no additional complications were observed in either approach.[17].

During reduction, the bowel is inspected for obstructions or atresia. Bowel anastomosis is usually not performed if the bowel is swollen. In cases of obvious bowel atresia, an end ostomy may be created to allow for feeding until the intestines recover. Bowel perfusion is closely monitored during the reduction process. Elevated intra-abdominal pressure has the potential to result in abdominal compartment syndrome, affecting organ function and causing complications.[14], [17]

The postoperative care of infants with Laparoschisis depends on the extent of intestinal injury. Nasogastric decompression and total parenteral nutrition (TPN) via a central venous catheter are typically required. Feeding is initiated once bowel function returns. Trophic feeds are gradually increased to improve peristalsis. Maternal expressed breast milk is beneficial if available. Early oral stimulation helps with feeding milestones. Erythromycin has no effect on achieving full enteral feeding after Laparoschisis repair. Prophylactic antibiotics should be discontinued within 48 hours of abdominal wall closure unless there are signs of infection. The frequency of necrotizing enterocolitis (NEC) in Laparoschisis is higher than expected but tends to be less severe[18]. Short bowel syndrome (SBS) can occur in complex cases with extensive bowel injury and may require prolonged parenteral nutrition. Management of SBS involves various interventions and a multidisciplinary team. Cryptorchidism is common in male infants with Laparoschisis but often resolves spontaneously within the first year. Watchful waiting is recommended, and laparoscopic orchidopexy is an option if needed[19] .

The short-term prognosis for infants with laparoschisis is heavily influenced by their bowel condition at birth. Simple laparoschisis cases typically have an average hospital stay of 41±32 days and a mortality rate of 3.4%. In contrast, complex laparoschisis cases involve a lengthier hospital stay averaging 85±60 days, and a higher mortality rate of 9.3%. Over 70% of infants with complex laparoschisis and roughly 25% of those with simple cases often require additional surgical interventions due to bowel obstruction stemming from issues like adhesions, anastomotic stricture, or volvulus. The Laparoschisis Prognostic Score (GPS) is a validated

scoring tool used to evaluate bowel necrosis, matting, atresia, and perforation shortly after birth, with the aim of predicting outcomes. In infants with complex laparoschisis, elevated urinary intestinal fatty-acid-binding protein (I-FABP) levels suggest intestinal mucosal damage. However, it's important to note that urinary I-FABP levels are not consistently reliable for predicting outcomes such as the initiation of feeding or the length of hospital stay. [20]

In the long term, infants with laparoschisis still face challenges in terms of growth and neurodevelopmental outcomes. Several factors contribute to these challenges, including prematurity, being small for gestational age (SGA), prolonged use of total parenteral nutrition (TPN), recurrent infections, and extended hospital stays. Additionally, there are concerns regarding the potential consequences of repeated exposure to anesthetic agents, particularly in instances necessitating multiple surgical procedures[21].

While follow-up studies on laparoschisis are somewhat limited, they offer valuable insights into long-term outcomes. In terms of growth, a substantial number of infants with laparoschisis encounter suboptimal weight gain during their initial two years of life. However, the majority of these infants exhibit "catch-up" growth and experience notable improvements in weight z-scores and growth percentiles during extended follow-up. It's worth noting that infants with complex laparoschisis typically have lower weight z-scores when compared to those with simple laparoschisis.[23].

Neurodevelopmental outcomes in laparoschisis patients at 1-2 years of follow-up seem to be on par with non-surgical neonatal intensive care unit graduates who share similar birth weight and gestational maturity[24]. Nevertheless, it's important to note that children with complex laparoschisis typically experience poorer neurodevelopmental outcomes in comparison to those with simpler cases[25].

During childhood and adolescence, laparoschisis patients commonly experience abdominal pain and recurrent gastrointestinal symptoms. These symptoms include difficulty with completing a meal, gas bloat, gastroesophageal reflux symptoms, and abdominal pain. Despite these challenges, studies have reported that the overall quality of life of laparoschisis patients, as reported by both patients and parents, is comparable to that of healthy individuals.[26]

Conclusion

In conclusion, laparoschisis is a congenital condition which is usually diagnosed before birth through ultrasound and can be further evaluated using MRI. The cause is not well understood.

Delivery plans depend on various factors, and vaginal delivery is generally possible.

Following birth, the primary objective is to reposition the organs within the abdomen while minimizing any potential injury. Postoperative care involves monitoring for complications and initiating feeding once bowel function returns.

Long-term outcomes vary depending on the severity, with complex cases experiencing longer hospital stays, higher mortality rates, and potential complications requiring additional surgeries.

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Competing interests

Authors have declared that no competing interests exist.

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