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Idiopathic colonic and gastric perforation in a full term newborn

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Abstract
Idiopathic Perforation of the transverse colon in neonate is a rare finding in clinical practice. We reported a case of Idiopathic neonatal colonic perforation, colic perforation is an extremely rare condition and very few cases have been reported in a full term new-born. We reported the first case of a new born at 39 weeks of gestation, who, twelve days after birth, had pneumoperitoneum without any demonstrable cause. Surgical exploration showed a transverse colonic perforation, the perforation was closed primarily. The postoperative course was simple, the following up was made every two weeks during six months. The second case Spontaneous neonatal gastric perforation is rare. We report the case of a full term 4-day old newborn who required resuscitation at birth. On the second day of life he had extensive abdominal distension. Abdominal x-ray without preparation showed pneumoperitoneum. Laparotomy showed a perforation at the level of the antrepyloric, measuring 1,5 cm in diameter, treated by simple surgical suture. The patient dead on the first post-operative day.

Keywords: Colic and gastric, idiopathic perforation, neonate term newborn

Introduction
The term idiopathic gastrointestinal perforation refers to any perforation of the gastrointestinal tract without an obvious cause [1]. Colonic involvement is rare, the majority of perforations being secondary, linked to ulcerative-necrotizing enterocolitis (ECUN), Hirschsprung's disease, anorectal malformations (MAR) and other causes of neonatal intestinal obstruction [2, 4]. Neonatal resuscitation is the most important history. The pathology is frequently described in premature babies with low birth weight. However, a few cases of full-term neonates have been described [5]. In the presence of early and appropriate diagnosis and management, the prognosis is good [4]. Spontaneous gastric perforation is rare in term neonates and accounts for 10 to 16% of neonatal gastrointestinal perforations [6]. It is a condition with a serious prognosis. The high mortality rate in these patients can be improved by early diagnosis and rapid resuscitation [7]. The usual age of onset of neonatal gastric perforation is between two and seven days and there is a predilection for the black race and the female gender [8]. Additionally, several risk factors are associated with the condition; prematurity, low birth weight, exchange transfusion, premature rupture of membranes, toxaemia of pregnancy, breech delivery, maternal diabetes, placenta previa, amniotic infection or Caesarean section [7, 8]. In the majority of cases, it requires early surgical management, which can range from surgical suture to partial or total gastrectomy. Since Siebold's first description in 1825 [8], more than 300 cases have been reported in the literature [9]. The rarity of this pathology in the world motivates this publication. The objective was to describe the clinical picture and the management of two cases of colonic and gastric perforations observed at the university clinics of Lubumbashi in September 2020.

Patients and observation
Our study concerns two observations of idiopathic neonatal colonic and gastric perforations treated in our pediatric surgery department of the university clinics of Lubumbashi. The first patient was a 12-day-old female newborn transferred from the pediatric

ward for abdominal bloating. At 10 days from the consultation, the patient presented with a fever, quickly followed by vomiting and cardiac arrest. gases and materials a day later. The parents allegedly gave him gout quinine and vitamin B6, in unreported doses. The absence of improvement in the symptoms would have led to hospitalization in a medical center for 8 days before being transferred to the university clinics of Lubumbashi where the patient was received in paediatrics. The diagnoses of intestinal sub-occlusion and sepsis complicated by hypoglycemia and moderate dehydration were retained. The patient was then transferred to surgery two days later, under treatment with Cefotaxime (300 mg intravenously twice daily), Ampicillin (200 mg intravenously twice daily) and Lactulose (2.5 ml Per Os per day). The mother is 25 years old, with no history of diabetes. The course of the pregnancy was done without any particular event and the prenatal consultations were only made in the third trimester. The delivery occurred at the 39th week of amenorrhea according to the date of the last menstrual period. The rupture of the membranes occurred 8 hours before the delivery, the newborn would not have been resuscitated and would have weighed 3000 grams, the meconium would have been emitted 4 hours after the delivery which took place in a rural medical center.

The newborn showed abdominal bloating that was diffuse and progressive onset with vomiting occurring 2 to 3 times a day, very abundant, greenish in appearance. There is a notion of incessant crying, the last stool would have been issued about 10 hours before the consultation. On physical examination, the salient elements are: a polypnea at 70 cycles per minute (other vital signs being within the norms), pale palpebral conjunctivae and a shiny, diffusely bloated abdomen, with increased visibility of the network venous under the skin, supple and defenceless, diffusely tympanic and silent on auscultation. Digital rectal examination revealed no contributing signs. The ultrasound identified a lot of digestive gas, the presence of a great abundance of liquid in the peritoneal cavity and the radiography showed an intrahepatic diaphragmatic gas crossing. After resuscitation, the patient was taken for an exploratory laparotomy which showed pneumoperitoneum, dilated intestines, about 30 cc of pus collected in the cul-de-sac of Douglas and between the intestines, fibrin almost everywhere in the large peritoneal cavity and a punctiform perforation located in the middle third of the transverse colon with intact edges, identified after digital adhesiolysis.

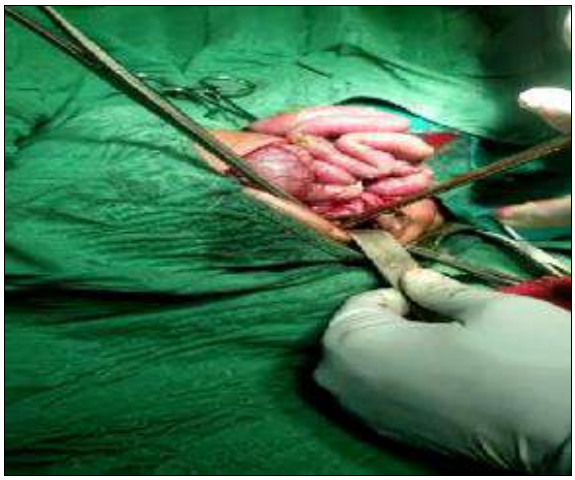

Fig 1: Colon perforation

After complete adhesiolysis, exploration of the rest of the abdominal cavity did not show any abnormality. Nothing was suggestive of congenital megacolon or any other mechanical cause of neonatal obstruction. The loops were emptied from below from the angle of Treitz to the rectum, followed by suturing of the purse-string perforation with 5/0 Vicryl and omentoplasty. The cavity was washed with 1 liter of physiological serum before the staged closure of the wall. In the postoperative period, the patient was cared for by a mixed team including resuscitators and surgeons. He provided antibiotic therapy (Cefotaxime 3x500mg), liquid infusion (Physiological serum, 5% glucose serum and Ringer Latacte serum, in total 1.5 L for 24 hours), parenteral analgesia (Paracetamol i suppository a reason of 2x 30mg).

The second patient, It was a 4-day-old female newborn, who was brought to the surgery department of the University Clinics of Lubumbashi on September 10, 2020 for abdominal bloating, crying. incessant and feverish. In the history, he was born at term in a local hospital with an APGAR score of 4 at the first minute and he was resuscitated for about ten minutes. He presented abdominal bloating, fever and incessant crying since the first day of life despite the emission of meconium. He was put on cefotaxime, ampicillin, and paracetamol. The persistence of abdominal bloating without transit disorder motivated his transfer to the university clinics of Lubumbashi. At the physical examination carried out at the university clinics of Lubumbashi, the anthropometric parameters were as follows: weight: 3950 g, cranial perimeter: 36cm, thoracic perimeter: 36cm, brachial perimeter: 12cm and height: 53cm. Somatic examination had shown integumentary pallor with skin recoloration time of less than 3 seconds, bloated abdomen, abdominal defense, with diffuse tenderness on palpation, tympanism, peristalsis was present and there was no organomegaly. Examination of the external genitalia revealed swelling of the vulvar region (Figure 2). In front of this picture, an abdominal X-ray without preparation had been carried out and had revealed a pneumoperitoneum. Ultrasound had shown a liver of normal volume, detached from the diaphragm by a hypoechoic collection with the appearance of a hypoechoic granule; the abdominal cavity filled with a hypoechoic collection, more abundant in the submesocolic region and compressing the handles visible in depth and not dilated. The diagnosis of neonatal peritonitis was retained and an exploratory laparotomy was decided.

one from a greenish-looking liquid with a sour smell was highlighted. The presence of papyraceous adhesion above mesocolic justified adhesiolysis by digitoclasia. Raising the anterior edge of the liver revealed an oval breach about 1.5 cm in diameter at the level of the antrepyloric part of the stomach.

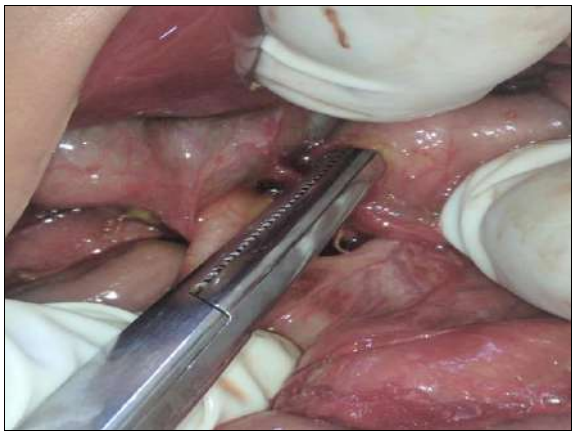

Fig 2: Gastric perforation

The aspiration of the liquid, the adhesiolysis, the suture in points separated from the breach followed by a seroserous plication (discharge suture) with vicryl N°2/0, the abundant cleaning of the abdominal cavity with saline solution and a drainage of the above-mesocolic level and of the Douglas and the abdominal wall suture had been made,

Fig 3: The abdominal wall suture

Postoperatively, the newborn was cared for by a mixed team involving paediatricians, resuscitators and surgeons. He had received antibiotic therapy consisting of ceftriaxone and gentamycin, a transfusion, a fluid infusion, analgesia and warming in a hot water bottle. He had died a day after the operation a picture of shock with respiratory distress.

Discussion

Since 1863 Breslau has defined spontaneous intestinal perforation as any perforation unrelated to ECUN, they are also called idiopathic, focal or localized perforations. They represent 30% of intestinal perforations occurring in newborns [10, 12]. About 15-30% of colonic perforations that occur in newborns and infants are idiopathic. Apart from ECUN, other causes of secondary perforation must of course be excluded such as Hirschsprung's disease, anorectal malformations, colon atresia, meconium plug syndrome, stercoral perforations and rarely, cystic fibrosis [3, 13]. The clinical presentation was neither compatible with ECUN nor with Hirschsprung's disease. Hematological investigations, bacteriology did not reveal germs and at laparotomy, no transition zone was identified, just as any other mechanical anomaly was not isolated. A biopsy, looking for Hirschsprung's disease should be done, even if clinical and laparotomy

Since the patient's follow-up did not refer to clinical elements suggesting congenital megacolon, this diagnosis can be excluded for the moment. Idiopathic perforation of the colon is a very rare entity. Unlike idiopathic intestinal perforations which are more frequent and whose risk factors are well identified (prematurity, low birth weight and neonatal asphyxia), there are still no clearly established factors for idiopathic colonic perforations [11, 14]. Some authors have highlighted traumatic factors such as rectal thermometer, barium enema or other rectal instruments [14]. Such manipulations were not done in the case of our patient. For newborns who suffered from neonatal asphyxia, Touloukioan *et al*. Attempt to explain perforation by the fact that anoxia or hypoxia can cause intestinal ischemia, with underlying NEC [15]. This is not applicable in our case where the patient was born at term and was not resuscitated, therefore, has no history of perinatal asphyxia. Weinberg *et al*. Have proposed a vascular theory: the perforation would occur because of ischemic necrosis, secondary to a localized vascular accident occurring in the intestinal wall. This theory results from the fact that in their series, 6 cases presented localized perforations on the antimesenteric border of the intestinal wall, which is the terminal point of the intestinal arterial network [16]. The fact that the perforation, in our case, is located antimesenteric could be linked to this theory, but we did not macroscopically identify an area of ischemia. Some still attribute.

Idiopathic colonic perforations to pseudo-obstruction of the left colon. It is an entity that occurs in newborns of diabetic mothers, where in response to hypoglycemic attacks the excess secretion of glucagon leads to a spasm of smooth muscles, thus causing a pseudo-occlusion [14]. Two facts lead us not to consider this theory for our case since the mother is not diabetic and at the laparotomy, the right and left colonists did not present any morphological abnormality.

Idiopathic colonic perforation is often difficult to diagnose clinically. In their series, Chang *et al*. Found fever to be the most common symptom. It is common to find vomiting, tachypnea, respiratory distress and abdominal bloating, which remains the most frequent sign [13, 17, 18]. Fever, vomiting, tachypnea and abdominal bloating were found in our case. However, none of these signs are pathognomonic for idiopathic colonic perforation. According to Soo-Hong Kim *et al*. The average duration between the first signs and hospitalization in a specialized environment is 6.7 ± 3.0 days. Our patient took 10 days to be admitted to our department. Radiography identifies pneumoperitoneum in more than 80% of cases. In the presence of massive pneumoperitoneum, gastric and colonic perforations should be considered. X-rays allow rapid diagnosis and appropriate surgical management [17]. In our case, the X-ray showed significant intestinal distention and abundant pneumoperitoneum. Ultrasound allowed us to rule out other causes of neonatal intestinal obstruction such as duplication and intestinal atresia. Surgical treatment, when undertaken in time, with minimal fecal contamination, can have a better prognosis. However, this treatment depends on 3 essential factors: the onset of symptoms, the degree of peritonitis and the general condition of the patient. In the past, it was the rule to perform a resection of the perforated portion and to perform a colostomy. Currently, primary closure or

Resection-anastomosis are performed for hemodynamically stable patients. However, many data suggest good results in case of primary closure alone, this technique is increasingly applied, sometimes by laparoscopy. For patients with severe peritoneal contamination and/or intestinal ischemia, two-stage surgical treatment after colostomy is more advisable [13, 17, 19]. After preoperative resuscitation, the patient was hemodynamically stable, allowing primary closure.

Contrary to generally accepted literature, recent studies find that the most frequent perforation sites are no longer the proximal colon, even less the vermiform appendix. It is now the transverse colon, the sigmoid and the spleno-colic angle that are

evoked [3, 4, 13, 17, 18]. As far as we are concerned, the transverse colon was the site of perforation. Thanks to early management, mortality linked to colonic perforations is significantly reduced [3, 18]. Our patient survived the surgery and did not present any complications in the postoperative period. Review 6 months after the operation, she is in perfect health. Idiopathic or primitive neonatal gastric perforation has been known since the first observation reported by Siebold in 1825. It accounts for 10 to 16% of neonatal gastrointestinal perforations [20]. Its mortality is heavy [20]. The usual age of onset is between two and seven days and there is a predilection for the black race and the male gender [21]. Abdelhalim Naji *et al* (2014) in Morocco reports a case of a 2-day-old newborn [22]; Giovanni DR *et al*l (2014) in India reported a case [20]; Mustafa Aydin *et al* (2015) report a case of a 2-day premature baby in Turkey [2]; Yong Hoon Cho *et al*l (2015) in Korea described an observation of 11 cases over 10 years, from January 2005 to December 2014, with an average gestational age of 29.7 ± 3.7 weeks of amenorrhea [21]. Our patient was a 3-day-old newborn, female and black. Several causes are put forward in the occurrence of the perforation. Thus, perforations of congenital origin by agenesis of the gastric musculature have been reported, causing lesions of the type of linear tear at the level of the lesser curvature; then perforations of ischemic origin (neonatal suffering, septic embolism), of mechanical origin (gastric distension after too much mask ventilation, gastric tube perforating the stomach), of drug origin (corticosteroids, indomethacin in the interauricular communication) or of functional origin (neurological affection, gastric atony, pyloric spasm in the event of neonatal stress) resulting in punctiform perforations of the anterior or posterior gastric wall.

In addition, several risk factors are associated with the condition: prematurity, low birth weight, exchange transfusion, premature rupture of membranes, and toxaemia of pregnancy, breech delivery, maternal diabetes, placenta previa, amniotic infection or caesarean section [21, 23]. Among the etiological factors listed in the literature, the factors found in our observation were: a depressed Apgar score, neonatal suffering, and resuscitation. In this pathology, two modes of presentation are described. The first is an array of pneumoperitoneum which is often associated with an array of shock. This table requires surgical treatment and its prognosis is pejorative; the second, a picture of massive digestive hemorrhage for which medical treatment may be sufficient [24]. In our case, it was a picture of pneumoperitoneum because the x-ray of the abdomen without preparation had revealed pneumoperitoneum. In addition, we did not objectivize digestive hemorrhage. On the therapeutic level, we resorted to a suture in separate points, a drainage of the supra mesocolic floor as well as the Douglas had been carried out. This treatment is described by several authors [21, 24]. For cases diagnosed and treated early, the evolution is favorable. However, the outcome is unfavorable for cases treated late [8]. In our case, the evolution resulted in death on the first postoperative day following the delay in management, multi-organ failure and a problem managing pediatric anesthesia.

Conclusion

Neonatal idiopathic colonic and gastric perforations are very rare entities, which also occur in term neonates with or without a history of resuscitation. Its risk factors are still discussed, they have no pathognomonic sign, but should still be sought even in the absence of significant pneumoperitoneum once ECUN has been ruled out. Early diagnosis associated with adequate surgical management significantly reduces mortality.

Conflicts of interest

The authors declare no conflicts of interest

Author's contribution

All authors have contributed to the conduct of this work and all authors declare to have read and approved the final version of this manuscript.

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