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Case Report

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Can Almost-Fully Gangrenous Midgut, Completely Survive After Closure of The Abdominal Cavity?

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Abstract

Here we are presenting a case of duplication cyst presented with almost fully gangrenous midgut in a neoborn secondary to a duplication cyst in the proximal jejunum, presented at Nizwa Hospital, Oman. Gastrointestinal (GI) duplications are rare congenital malformations that may vary greatly in presentation, size, location, and symptoms [1]. GI duplications may present as solid or cystic swelling, intussusception, perforation, or GI bleeding or very rare volvulus. In our case a neoborn presented with intestinal obstruction and investigations pointed to intestinal obstruction due to duplication cyst. Laparotomy findings showed near total midgut volvulus causing strangulation with subsequent almost fully gangrenous small bowel and a large duplication cyst at the proximal jejunum. Bowel colour did not improve after about one hour of de-rotation, warm fomentation and increase O2 supply. Depending on specific criteria we resected only the duplication cyst and we did primary intestinal anastomosis of that ischemic bowel. Abdomen was closed without a drain. We put specific parameters for post-operative observation. Was explained to parents that we will observe and may need re-laparotomy after 48- 72 hours if there is deterioration of the vitals, deranged investigation parameters and/or worsening of general condition of the baby. Baby improved and bowel survived. Barium meal follow through after one year showed normal bowel distribution and peristalsis. This is the first case report in such pathology and may change the view of management of near fully gangrenous bowel not improving with intraoperative resuscitation.

Discussion: *We reviewed articles on duplication cyst and its complications, Also articles in midbowel gangrene and its management and the prognosis.*

Conclusion: *Can criteria of bowel gangrene which indicate resection, change?*

Keywords: duplication cyst, intestinal volvulus, midgut gangrene, intestinal obstruction, strangulation.

Case report

The period managed by Neonatology team

A post term, 41 weeks and 2 days, male baby. Mother is diabetic and g3p2. Baby was born via lower segment caesarean section. Apgar score 3/1, 6/5, 8/10 minutes. Birth weight 3.62 kg. When the baby was delivered, he was flat, cyanotic, with heart rate <100/mt. Oral suction and tactile stimulation were done and baby started to take gasping breath. Nasal suction and ambu-bagging were done and he was settled and became active and pinkish. Baby was then shifted to maternal side. At 7 hours of life, he became hypoglycaemic for two times, (blood sugar 2.4 mmol) and was corrected. Baby was on breast feeding. At 12 hours of life baby started to have vomiting of dark green to blackish material. Nasogastric tube was inserted, aspiration showed the same colour with brownish admixture? Blood. Baby was shifted to NICU (Neonatal intensive care unit). On examination: afebrile, awake, with fair activity, dehydrated with mild tachypnea. Reflo: 4.8 mmol. BP 91/51 mmHg. HR: 134/mt RR: 76/mt Weight: 3.73Kg. Jaundice+. Abdomen:

Mild distension, soft. Bilateral scrotal swelling (hydrocele). The baby was resuscitated and kept nothing per mouth. IVF. Boluses of normal saline. NGT in free drainage and hourly aspiration with replacement of the output. *Abdominal x ray*: Not impressive (like normal) [Figure 1]. *Abdominal ultrasound*: at 16 hours of life: Slight turbid urine in the urinary bladder. Mild sub hepatic and pelvic free fluid with impression of unremarkable study. So the baby was not referred to Pediatric Surgery opinion. Again he had vomiting brownish blood and with melena. Then at 36 hours of age was referred to Paediatric Surgeons.

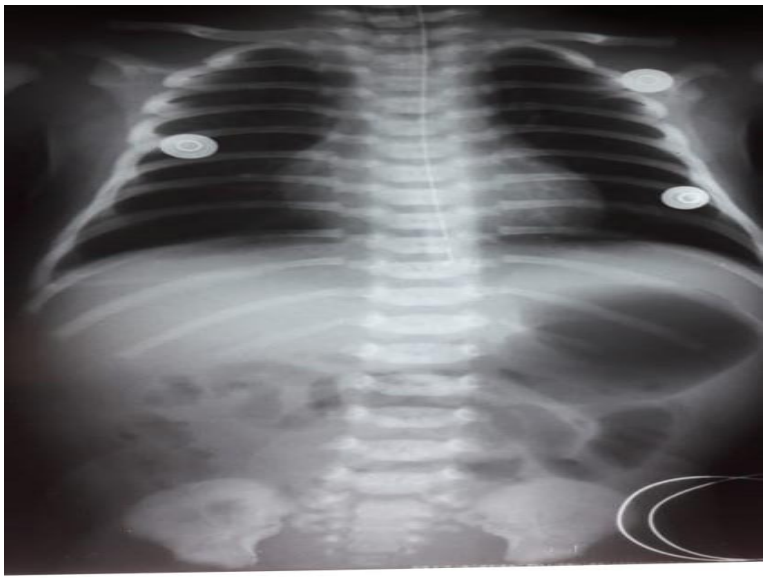

Figure 1: First XR taken after 6hr of birth. Looks normal abdominal XR.

Management by The Pediatric Surgery team

The baby was; vitally stable, pink, mildly dehydrated. Abdomen; mild distension, soft with palpable loops at right side of the abdomen, no palpable mass. Repeated abdominal XR (fig. 2): Few significantly dilated loops at upper abdomen.

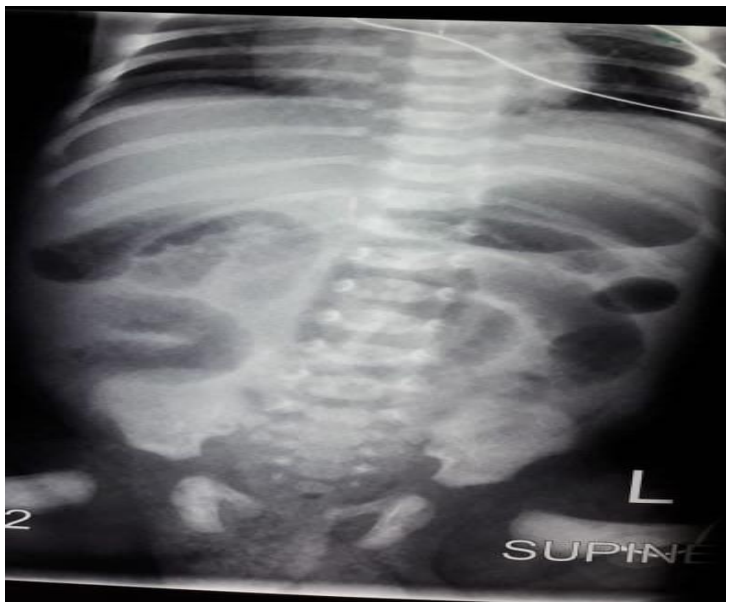

Figure 2: AXR: Few significantly dilated loops at upper abdomen.

Abdominal ultrasound was repeated and repeated as Mild free fluid level intra peritoneal. Dilated small bowel loops measures 11 mm, with thick oedematous wall (3 mm). Collapsed large bowel. Intestinal obstruction is suspected. Cystic structure is seen in the right iliac fossa with thick wall and turbid fluid contents, measures 35x25 mm. Differential diagnosis; Enteric duplication cyst ??Mesenteric cyst. Exploratory laparotomy (48 hours after birth) through a right lower transverse muscle cutting incision the findings were; Bloody peritoneal fluid, moderate amount. Proximal Jejunal duplication cyst about 5x4 cm. at the proximal Jejunum 70 cm from the DJ flexure. Volvulus of all the midgut distal to the duplication cyst till 4 cm to the ileocecal valve. That bowel was gangrenous nearly dead black. With 4 twists

at the root of its mesentery. Enlarged mesenteric lymph nodes. No evidence of malrotation with normal position of DJ flexure at the left of the midline and caecum at the right lower quadrant of the abdomen and no Ladd's bands. The bowel was untwisted. Warm fomentations were applied and ventilator oxygen was increased. After one hour still no much changes except: a) arterial pulsation of the mesenteric arteries started to appear. b) The colour was still dark but spots of pinkish discolouration appeared (Figure3). Then decided to resect the duplication cyst (Figure4). c) There was little venous blood ooze, no arterial bleeding at the cut edges. d) Weak or no clear peristalsis. But the bowel doesn't look mummified or 100% dead on benching it. Depending on the above observational criteria, it was decided to do primary anastomosis in one layer interrupted sutures using 4/0 vicryl and the bowel was returned back to the abdominal cavity and the abdominal wall was closed without drain. It was planned to monitor: 1) Vital signs, 2) laboratory investigations as blood gases, CRP (C reactive protein). Haemoglobin, Platelet count, Total and differential white cell count, RFT (renal function test). LFT (liver function test). Daily and if there is deterioration of vitals or general condition. 3) Urine output and Bowel output through the nasogastric Tube and per anal. 4) Frequent abdominal examination. Everything was explained to parents with drawing on papers. Possibility of re-laparotomy and excision of dead bowel in case of deterioration and even possibility of death was explained either during surgery or post-operative. Consent was taken. Post-operative period was uneventful, better than expected. We electively, delayed the oral intake for 7 days to avoid paralytic ileus of the exhausted bowel. NGT output was reducing in volume and clearing in colour after 48 hours. First the baby passed dark blood then passed green meconium at third post-operative day. Vitals were stable. Blood investigations including CRP (C reactive Protein) were normalizing. Started progressive NGT feeding at 7th post-operative day. And baby was discharged at 13th day post-operative in good condition. Followed up in the OPD and found to be doing well, gaining weight and normal bowel habits. At one year of age barium meal follow through was normal. (Figure5).

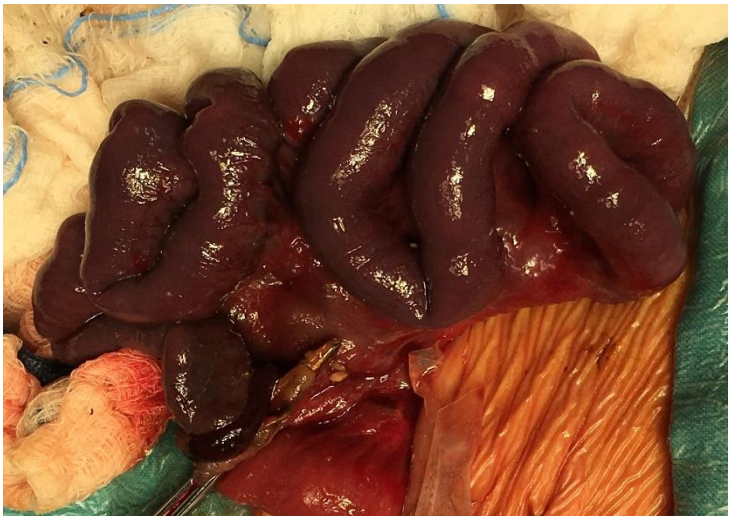

Figure3: Midgut after one hour of resuscitation and excision of the duplication cyst.

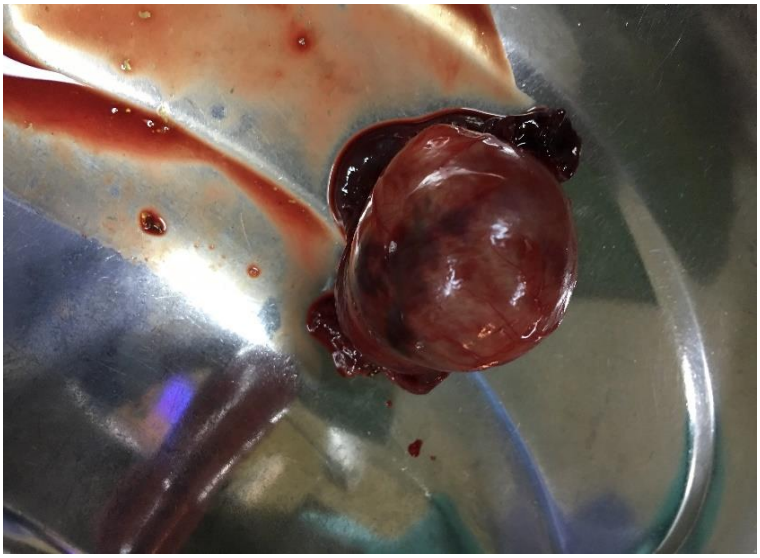

Figure 4: Excised duplication cyst.

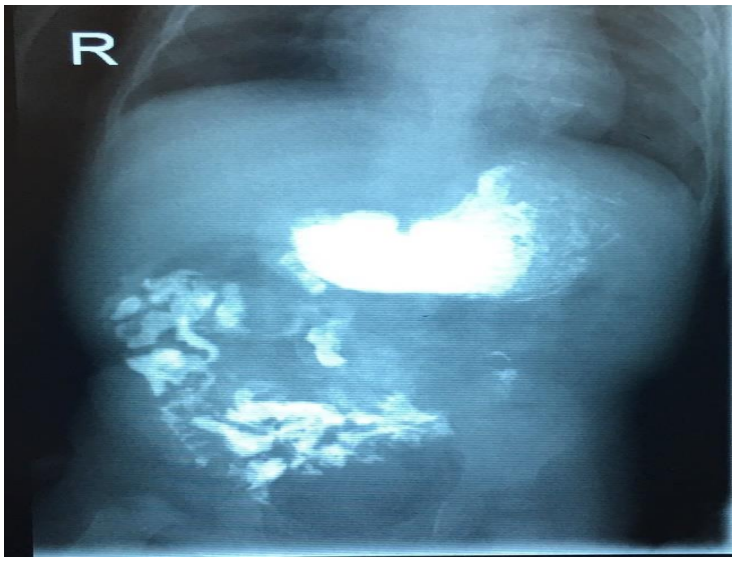

Figure 5: Barium meal after one year of surgery.

Discussion

Gastrointestinal (GI) duplications are rare congenital malformations that may vary greatly in presentation, size, location, and symptoms [1]. They consist of foregut duplication cysts, small bowel duplication cysts, and large bowel duplication cysts. Foregut duplication cysts are categorized on the basis of their embryonic origin into oesophageal, bronchogenic, and neuroenteric cysts [2]. Small bowel duplication cysts can be associated with all three small bowel subtypes: Duodenal, Jejunal, and Ileal. Jejunal duplications are the most common, followed by Ileal and duodenal duplications [14]. Duodenal duplication cysts makeup 2-12% [15]. Ileal duplication cysts makeup about 44% [16]. Jejunal duplication cysts makeup about 50% of GI tract duplications [17]. Duplication cysts can also be cystic (80%) or tubular (20%) [18]. The definitive pre-operative diagnosis of a duplication can be difficult [3]. The diagnosis can be achieved following a high level of clinical suspicion especially with the sonographic evidence of an intra-abdominal cystic swelling with peristaltic muscular contractions of the cystic wall [4,6]. The main differential diagnosis in neonates is intestinal atresia in which the gross proximal dilatation may mimic a cystic lesion on ultrasonography [5,6]. Malrotation and Meckel's diverticulum are typical in Infants (< 1year old). Hirschsprung's disease will fit into both categories. Antenatal diagnosis is advancing with increasing clinical experience [6]. In cases of Intussusception, perforation, or GI bleeding, a high index of suspicion is required in such cases. The mass effect of the duplication cyst would precipitate a volvulus [7]. About 75 % of all midgut volvulus cases occur within the first year of life, the majority of which are in the first month [8,9]. Mid-gut volvulus without malrotation causing intestinal obstruction in neonates is not a common finding [10]. Prognosis of midgut volvulus in neonates and infants younger than 1 year remains poor, as diagnostic findings may not be apparent until gut infarction had occurred [11]. As in our case of volvulus. Severe bowel loss and intestinal failure result in long periods of hospitalization and prolonged total parenteral nutrition. With its associated substantial morbidity, which includes repeated episodes of septicaemia, derangement of liver function and frequent admission to the hospital [12]. Intestinal and/or liver transplantation may be required. The quality of life of these patients and their family can be poor [13]. In an article was written on Indications and outcome of childhood preventable bowel resections in a developing country. There were 22 preventable bowel resections. Only one of them was midgut volvulus. Seven patients died (31.8% mortality). The midgut volvulus was one of the dead cases. The conclusion of that article was; there is a high rate of morbidity and mortality in these cases of preventable bowel resection [19].

In our case a proximal Jejunal duplication cyst is the cause of midgut volvulus and the subsequent strangulation of the bowel which is very rare. It is known in management of strangulated bowel, if the colour not improved to near normal, either the bowel will be resected or kept in the abdominal cavity with keeping the abdominal wall opened for second look. In our case, the bowel colour not significantly improved and in spite of this, we did resection of the duplication cyst, and primary anastomosis of the ischemic intestine. The bowel was returned back to the abdominal cavity and the abdominal wall closed without a drain. In spite of doing primary anastomosis in ischemic bowel, the healing was perfect and no leak or stricture.

Conclusion

This is the first case to do primary anastomosis in ischemic bowel and to return back to the abdominal cavity, of almost fully gangrenous midgut while still dark in colour after long time of resuscitation (fig.3). And in spite of this closure of the abdomen, with no drain and the result is full survival of the intestine. The criteria which were used in taking the decision to close the abdominal cavity, and which were for the first time to be used in such

highly compromised, ischemic bowel: a) arterial pulsation of the mesenteric arteries started to be felt. b) The colour still dark but spots of pinkish discolouration appeared (Figure1). c) Little venous blood ooze, at the cut edges of the intestine, no need to wait for arterial bleeding. d) Weak or no clear peristalsis with the bowel doesn't look mummified or not 100% dead on benching it. The use of post-operative monitoring method which is commonly used in critical patients. Full counselling to the parents in all points is a must. The success of this case deserves applying these criteria in severely ischemic bowel due to strangulation. Because removal of the affected midgut will end in short bowel syndrome with its known morbidity and mortality. Also keeping the bowel in the abdominal cavity without full closure will add morbidity and will prevent the trial of keeping it in a full natural environment for survival, which I think was the main factor for the bowel to regain its viability in our case. We need more cases to prove these criteria in such conditions.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflict of interest

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