A MISSED SPONTANEOUS SPLENIC RUPTURE IN A NIGERIAN NEONATE – A RARE AND CHALLENGING CASE REPORT AND REVIEW OF LITERATURE.

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ABSTRACT

Background: **Splenic rupture is relatively uncommon in newborns but could occur from birth trauma, increase intra-abdominal pressure such as polyhydramnios and fetal ascites. Abdominal ultrasonography remains the radiologic investigation of choice in this clime.**
We present this rare finding, first in the literature from sub-Saharan Africa, to raise awareness, and improve our index of suspicion of this extremely fatal condition.

Case summary: **A 2-day old male who was delivered via elective caesarian section at 39 weeks gestational age. He cried well at birth but was noticed to have a distended abdomen and respiratory distress 4 hours after birth with a reduction of the packed cell volume from 40% at 5 hours of life to 23% on the second day after birth. Abdominal ultrasound scan showed hepatosplenomegaly with moderate ascites.**
An initial diagnosis of malrotation with midgut volvulus was made. Exploratory laparotomy was done on the 36th hour of life after optimization of the hematocrit level with intra-operative findings of a 3cm sub capsular splenic laceration and 100 mls of haemoperitoneum with clots. The bleeding point was identified and a haemostatic agent applied. Six hours post-surgery, he suffered a cardiopulmonary arrest and all efforts at resuscitation were unsuccessful.

Conclusion: **Management of this condition may be non-operative or surgical. Irrespective of the treatment approach, it is associated with high mortality in the setting of delayed diagnosis and intervention. Thus, it is important to have a high index of suspicion on any neonate with the triad of worsening anaemia, abdominal distension, and shock.**

Keywords: Birth trauma, Neonate, Nigerian, Splenic rupture

INTRODUCTION

Birth trauma in newborns is well described in the literature with pooled prevalence of 0.48% to 16.7%.[1] Intra-abdominal injuries occur infrequently in neonates and are often fraught with delay in diagnosis.[2] However, the suspicion of damage to the integrity of an intra-abdominal viscera should be raised in a neonate with a diagnosis of a hypovolemic shock and an intra-abdominal mass.[2]

Splenic rupture is relatively a rare and life-threatening situation in the newborn. The classic triad of this condition is hypovolemic shock, anaemia, and abdominal distension.[3] The diagnosis is a dilemma for the physician and denotes a surgical emergency that may lead to mortality if there is delayed intervention. Thus, it is imperative for not just the clinician, but also the midwives, to be armed with the possible symptoms that may manifest for early diagnosis and intervention to increase the chances of survival in such a neonate.[4] High index of suspicion and improvement in diagnostic tools like ultrasonography and computed tomography scans are important armamentarium in establishing early diagnosis.[5]

Splenectomy is no longer the standard treatment of splenic rupture in neonates as it increases the chances of post-splenectomy sepsis, however treatment should aim for hemostasis and preservation of the spleen.[5]

We therefore present this rare condition, first to be documented in the literature from sub-Saharan Africa, to raise awareness, and improve our index of suspicion of this extremely fatal condition.

CASE PRESENTATION

A 32-year-old para 2^{+1} (2 alive) woman, with 2 previous caesarian scars, delivered a 3.1 kg male neonate via elective caesarian section at 39 weeks gestational age.

Index pregnancy was booked, with a third trimester obstetric ultrasound scan findings of moderate fetal ascites and polyhydramnious. Mother claimed there was no fall nor abdominal trauma prior to this time.

The newborn was attended to by the midwives at the secondary facility and he cried well at birth without aggressive resuscitation, but was noticed to have a distended abdomen and respiratory distress 4 hours after birth. He was subsequently placed on intranasal oxygen at 1 L/min and intravenous fluids.

Initial full blood count done 5 hours after birth, showed a packed cell volume of 40%, white blood cell count of 13,800/mm^3, and platelet counts within normal range. Other biochemical analyses were also normal. Abdominal ultrasound scan done on the first day of life showed hepatosplenomegaly with moderate ascites.

By the next day, the baby was noted to be deteriorating, evident by poor urinary output, worsening respiration and progressive abdominal distention, necessitating a paediatric surgical review. Prior to the review a plain abdominal radiograph was done revealing a distended abdomen, and stomach displaced inferiorly with widespread opacity in keeping with a ground-glass appearance of an intrabdominal collection (Fig. 1), necessitating a paediatric surgical review.

Following the surgical review, it was noted that he had not yet moved his bowel. He had a nasogastric tube passed to decompress the stomach and bowel, and an abdominal paracentesis was done which showed a free flowing non-clotted blood.

An initial diagnosis of malrotation with complicated midgut volvulus was made. A repeat packed cell volume done on the 2nd day after birth (31 hours of life) has dropped to 23%. He was subsequently transfused with 60 ml of fresh whole blood and worked up for an emergency exploratory laparotomy after being adequately resuscitated. Intra-operative findings at surgery done on the 36th hour of life were; a 3cm sub capsular splenic laceration, grossly normal liver, bowel, and 100 mls of haemoperitoneum with clots (Fig. 2). The bleeding point was identified and a haemostatic agent (Surgicel) applied. The patient subsequently had another 60 mls of whole blood transfused intraoperatively with evacuation of the haemoperitoneum. The duration of surgery was 50 minutes, but patient had delayed recovery from anaesthesia necessitating manual ventilation with Ayres T-piece in the postoperative period because there was

Figure 1: Plain abdominal X-ray showing a stomach displaced inferiorly with a ground-glass appearance.

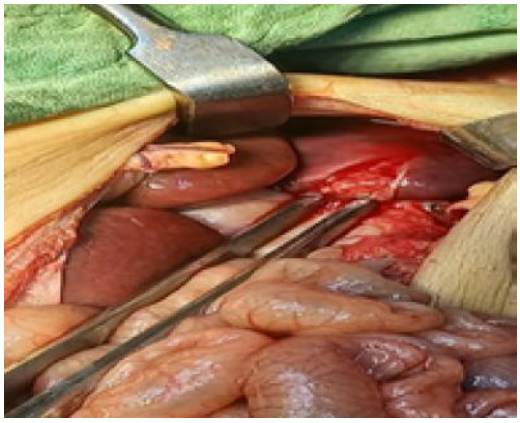

Figure 2: Supraumbilical transverse laparotomy incision with intra-operative findings of normal liver, bowel (no evidence of mal-rotation), and a grade II Splenic injury - a 3cm sub capsular laceration with hemoperitoneum.

no functional neonatal ventilator for mechanical ventilatory therapy. Six hours post-surgery, he suffered a cardiopulmonary arrest and all efforts at resuscitation were unsuccessful. A final peri-operative diagnosis of Grade II Splenic injury with significant hemodynamic instability was made.

DISCUSSION

Haemoperitoneum, in the newborn is a rare occurrence with less than 50 case reports in the world's literature making the true incidence of neonatal splenic rupture unknown, [4] and there are more autopsy diagnoses reported than surviving cases.[2, 5]

When it occurs, the presentations are variable and can be due to either of the several etiologies. Possible causes of haemoperitoneum in these age group could range from birth trauma, iatrogenic – during laparotomy for another condition, pathologic causes predisposing to splenomegaly or fragility, and small capsular lacerations to disintegration or shattering of the spleen from injury.[2] A high index of suspicion with the classic presentation of bleeding, abdominal distension, and hypovolemic shock, and improvement in diagnostic tools like ultrasonography, and computed tomography scan are important contributions in early diagnosis so that appropriate treatment can be implemented. Splenectomy has been the most preferred option of management of splenic ruptures, due to the most dreaded complication of post splenectomy sepsis, most paediatric surgeons now opt for spleen sparing surgeries.[2] Irrespective of the option of management, splenic rupture is associated with high mortality, thus, a high index of suspicion is needed to give the neonate a chance at survival.[2]

Various etiologies have been implicated and are linked to excessive compression of intra-abdominal cavity, thus birth trauma is the most implicated risk factor.[1] Trauma, likely from breech or complicated vertex vaginal delivery, and difficult caesarian breech delivery may result in excessive compression of the abdominal cavity. The index patient had polyhydramnios and moderate fetal ascites. These may cause difficulty in delivering the fetus during Caesarean section, and the fetal ascites can cause increase in intra-abdominal cavity pressure.[5] Aggressive newborn resuscitation also has the tendency of compressing the chest or abdominal capacity with subsequent blunt splenic injury, though this was not done in this patient.[5] Maternal fall or trauma, resulting in a precipitant delivery, has also been associated with fatal splenic haemorrhage in the newborn.[6] Some may occur spontaneously, just like in this index patient where no etiology from trauma was established.[7] Underlying clotting defects have been identified as a cause of significant haemoperitoneum as problems of vitamin K-dependent clotting factors have been associated with splenomegaly or fragility leading to excessive bleeding from minor splenic injuries.[3,8] Inherited coagulation disorders, disseminated intravascular coagulation from sepsis, and immune-mediated thrombocytopenia are other possibilities.[7] Erythroblastosis fetalis and congenital syphilis have been implicated as causes of neonatal intra-abdominal bleeding due to the increased fragility of an enlarged spleen.[5, 8]

The pathogenesis of neonatal splenic rupture is not well understood. However, it is thought that the mechanism of injury may be related to increased tension on the supporting ligaments of the liver and spleen that results from increased intra-thoracic pressure forcing the organs out of their normal positions.[5]

Splenic rupture occurs in two stages; initial sub capsular hematoma formation which may have only mild symptoms of unexplained anaemia and a left upper quadrant mass.[2] The second stage may occur after hours or days and is usually characterized by the rapid development of shock, as the spleen and mesentery tend to bleed rapidly and significantly.[3,4,5,6] This delayed presentation of splenic rupture is described mostly with primary hilar and/or parenchymal injury. The clinical presentation of ruptured spleen in neonates occurs within 14 - 21 days in 96% of cases as described by Longoboardi and colleagues.[9] However in this index patient, the presentation was early within 24 hours and rapidly deteriorating.

The classic triad of anaemia, progressive abdominal distension with hemoperitoneum as seen in this patient, should raise suspicion. Other complaints that may be present are complications that may arise from the triad, which include poor feeding, listlessness, rapid respiration, among others. Signs that might be seen include, pallor, tachypnea, tachycardia, a palpable left upper quadrant mass, and reduced urinary output amongst others .[2]

A full blood count may reveal low values of haematocrit, and a diagnostic paracentesis may reveal free-flowing blood. The most preferred diagnostic tool is the abdominal ultrasound or a computed tomography (CT) scan which may reveal an intra-abdominal free fluid. Ultrasonography is considered the most appropriate screening method in both traumatic and non-trauma situation, as it may illustrate the anatomy better than CT scan in some instances.[2, 10] This is not surprising as children are at higher risk for developing cancer caused by ionizing radiation compared to adults due mainly to the increased radio

sensitivity and a longer lifespan after exposure, hence adherence to the ALARA (as low as reasonably achievable) principle continues to be an important guide for all physicians in paediatric care.[10] Ultrasonography is operator dependent, and need skilled and experienced sonologist for identification of injuries in this subset of population however, it is more readily available and the onion-skin appearance of the intra-abdominal haematoma on ultrasound suggests a more chronic onset.[2] Abdominal x-ray may also be done which would show a mass effect and displacement of the stomach and bowel with ground-glass appearance as seen in this index patient. Other tests required are coagulation studies to rule out coagulation disorders.

It is imperative to resuscitate any newborn with suspected splenic rupture. Thus, fluids with transfusion of packed red cells should be commenced immediately while clotting factor deficiencies are corrected if present. Options of care are dependent on how haemodynamically stable the patient is. In an haemodynamically stable neonate, correction of any underlying coagulation abnormalities, blood transfusion, very careful serial critical care and observation of vital signs often can be applied successfully.[7] For those who are haemodynamically unstable, an exploratory laparotomy with or without splenic conserving surgery is indicated.[2] Splenectomy is done in the case of a shattered spleen, while splenic laceration can be repaired with mattress suturing, and application of haemostatic agents such as gelatin foams, oxidized cellulose, fibrin sealants, cyanoacrylate adhesives to control bleeding.[5] In this index patient, a Surgicel haemostat was placed into the area of the laceration. Post-splenectomy prophylaxis against encapsulated organisms such as *H.* influenza, *N.* meningitides, and *S.* pneumonia, is required in those who have had splenectomy to prevent the feared complication of overwhelming post-splenectomy sepsis (OPSI).

Prognosis is good only if detected early and definitive intervention instituted promptly, as splenic rupture in neonates is associated with high mortality.[5]

From the literature search done, this is the first case described in sub-Saharan Africa, hence the low index of suspicion at presentation which led to an initial wrong diagnosis, delayed intervention time, and eventual mortality similar with majority of earlier reports.[5]

CONCLUSION

Splenic rupture in a newborn is a rare condition and almost fatal in outcome if not promptly identified and treated. The aetiology could be related to birth trauma in many instances, but in some subsets the cause remains unknown. Irrespective of the treatment approach, it is associated with high mortality in the setting of delayed diagnosis and intervention as observed in this index case. Due to the uncommon incidence of splenic rupture, a high index of suspicion should be raised in a neonate with a palpable intra-abdominal mass, rapid decline in haematocrit level, and hypovolemic shock.

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