Niger J Paed 2013; 40 (1) :82 –84

CASE REPORT

Caecal duplication in a neonate causing intestinal obstruction

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DOI:http://dx.doi.org/10.4314/njp.v40i1.16

Accepted: 7th March 2012

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Abstract Intestinal duplication is one of the rare congenital abnormalities encountered. Duplications found on the small intestine are more commonly encountered in clinical practice than the large intestine and majority of these occurs in the ileum. We present a three-week old neonate with features of intestinal obstruction a presumptive diagnosis of intususception with presumed sepsis was made. Intraoperative findings revealed caecal duplication causing the intestinal obstruction. Limited excision and ileo-ascending colic anastomosis was carried out. Patient responded well and was discharged on follow-up.

Key words: Neonate, caecum, duplication, obstruction

Introduction

Congenital abnormality of the gastrointestinal system has been reported by Ambe *et al* [1] to be the commonest congenital abnormalities seen in neonate in North-Eastern Nigeria. Despite this, enteric duplications (ED) are rare congenital abnormalities [2] and have not been reported in recent times in North-Eastern Nigeria. Despite the rarity of this condition, many cases have been reported in literatures both within and outside Nigeria. [2, 3, 4, 5] Duplications in the small intestine are the most common and majority of these occurs in the ileum. [2] The duplication may either be cystic or tubular and most of them are located on the mesentry of the intestine. [3] Patients with colonic duplication who remain undiagnosed into adulthood, malignant changes into adenocarcinoma can occur. [2, 4]

Many theories of embryogenic origin to explain the cause of ED have been proposed, [2, 6] the split notochord theory, is believed to be due to errors in normal embryologic canalization, as the gut endoderm herniates through the notochord the developing gut become entrapped diverticular. While the persistent neuroenteric canal explains the vertebral anormalies.[5, 6] The embryonic diverticular theory is due to failure of regression of the embryonic diverticular this result in the small cystic duplications noted in the intestinal wall and sometimes enteric cyst seen in the presacral areas. [2,6] The theory of median septum formation, suggest that the walls of adjacent fetal bowel may be flattened by extrinsic compression with subsequent adherence and fusion resulting in doubling of the lumen. [6] The theory of partial twinning which suggest that the axial structures in hindgut duplications are twinned because of split in the primitive streaks, resulting in two notochords separated at their caudal ends which later fuses during cranial elongation of the embryo. [6] Finally, the theory of errors of epithelial recanalization that allow formation of cysts located intramural this would explain the presence of small sub mucosal duplications.[2, 6]

Despite these theories the exact etiology of ED has not yet been established. Signs and symptoms due to ED may be variable and simulate other diseases depending on their locations.[4,5,6] These varied signs and symptoms include; pain, vomiting, nausea, bleeding, abdominal distention with or without mass, dysphagia, dyspepsia, respiratory distress, perforation, chronic constipation and obstruction of the alimentary tract.[3-5, 7, 8] Most cases of ED are diagnosed incidentally especially at surgery.[2, 3, 5]

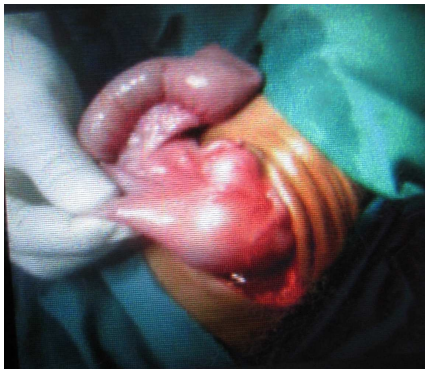
Plain abdominal radiograph and contrast radiographic studies such as swallow, meal, follow through and enemas could be of diagnostic value. [5] Ultrasound and magnetic resonance imaging (MRI) findings may be valuable diagnostic tools. [2,5] Computerized tomography scan (CT) is of use in delineating surrounding structures.[2, 5] Once the diagnosis is established, the treatment is surgical and the outcome is generally good. We present our experience in the management of three-week old neonate with caecal duplication.

Case Report

A.Y.U., a three week old male neonate presented to the Special Care Baby Unit of the Federal Medical Centre Yola on 18th June 2011 with abdominal distention, fever, vomiting and constipation for a week. Child had passed meconium within the first 24 hours of delivery. The antenatal history of the mother suggested no evidence of polyhydramnous, diabetes or hypertension. Delivery was at term attended by traditional birth attendant (TBA) but was uneventful. Upon examination, the significant findings were fever with temperature 38.5°C, the weight was 3.25 kg, the abdomen was uniformly distended with a mass measuring about 6 cm x 6 cm in the right flank with hyperactive bowel sound. A presumptive diagnosis of intususception with sepsis was made.

Plain abdominal radiography showed distended bowel loops with no air-fluid levels, but abdominal ultrasound scan was reported as normal. Blood culture yielded no bacterial growth. Complete blood count, differentials counts, Serum electrolytes and urea were within normal limits. The patient was commenced on antibiotics for presumed sepsis. Temperature became normal within three days of treatment, but there was no significant improvement with abdominal distention. On the fifth day abdominal distention worsened with visible peristaltic waves from right to left across the lower abdomen. Ten days into admission patient was optimized for exploratory laparatomy, which showed duplication of the caecum (Fig 1). Limited excision and ileo-ascending colic anastomosis was done, histological examination confirmed the diagnosis of caecal duplication. Patient responded to treatment and was discharged on followed-up.

Fig 1: Cystic duplication of the caecum



Discussion

The signs and symptoms of ED depend on the size, location and the nature of the mucosal lining, associated anomalies and complications. [5, 7, 8] However, most cases reported in literature present within the first year of life.[3-5, 7] Some patient may not be symptomatic until school age or adulthood.[3, 5, 8] Our patient is a male which agrees with many series reported in literatures, [2-5, 8] however other workers have reported no sex predilection, [9] while others found female: male ration of 9:4. [10] Patients may present with abdominal pain, this was observed with difficulties in our patient as he was having intermittent cry which may be due to abdominal pain. Vomiting was one of the main presenting symptoms in our patient, together with abdominal distention and palpable mass in the abdomen. These presentations were in consonance with other reports especially from Ile Ife and Ilorin. [2, 3, 5, 8] The index patient also had fever, which may be as a result of sepsis being one of the complications of intestinal obstruction because of bacterial translocation.[8] Diagnosis of ED is rarely made on clinical ground until surgery because of non specificity of symptoms and signs.[5] The index patient had localized type of ceacal duplication which may explain why there were no other congenital abnormalities especially of the skeletal system found on plain radiograph or ultrasound.[2]

Never the less other workers within and outside Nigeria have reported associated malformation which include; ventricular septal defect, malrotation of the gut, situs inversus, oesophageal atresis, Down syndrome, imperforate anus, mesenteric cyst, absence of the left rectus abdominis muscle and short ascending colon.[2-4, 8, 11] Abdominal ultrasound in this patient did not show any remarkable findings, there was a similar experience in Ilorin.[5] The diagnosis was missed on ultrasound may be because of low index of suspicion, limitation imposed by overlying bowel gas and probably the technical know-how of the sonographer. Computerized tomographic scan (CT scan) and MRI could have been very useful in establishing the diagnosis, which will show a three-layered image representing the duplication cyst, common wall and outer bowel wall.[2, 5] These imaging modalities have helped in making prenatal diagnosis in some cases.

However we lack the facilities to carry out these investigations. Laporatomy was carried out and the patient had limited excision and ileo-ascending colic anastomosis. Surgical treatment generally is offered to all patients with ED, the main considerations are the age, location, types of lesions and the condition of the patient.[3-5] Index patient did well postoperatively and was discharged on follow-up.

Although rare, enteric duplications do exist in our environment and this demand for high index of suspicion and meticulous management, to avoid patient's untold suffering and complications.

Conflict of interest: None
Funding: None

Acknowledgment

We thank the Nursing Staff in the Special Care Baby Unit, for the care given to the patient.

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