Case Report

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Duplication cyst of ascending colon presenting as an ileal volvulus in a child: A case report and review of literature

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ABSTRACT

Alimentary tract duplications are uncommon congenital anomalies that may be found anywhere along the gastrointestinal tract. They have a diverse presentation and often times present with complications of intestinal obstruction and/or lower gastrointestinal haemorrhage. We report a very rare finding of a colonic duplication cyst which presented as an ileal volvulus in a young girl. While available investigations were non-specific, the child survived on account of an early decision to operate. She had resection of the duplication cyst and ileo-colic anastomosis. She made good recovery post-operatively. A high index of suspicion is necessary on the part of clinicians to recognise this condition. Early operative intervention is necessary in the developing world setting where state of the art investigations are not available for accurate pre-operative diagnosis.

Key words: Congenital anomalies, colonic duplication cysts, complications, ileo-colic anastomosis, volvulus

INTRODUCTION

Alimentary tract duplications are uncommon congenital anomalies that may be found anywhere along the gastrointestinal tract from the mouth to the anus.[1] The incidence has been reported to be 1 in 4500.[1] More than 80% of cases present before the age of 2 years as an acute abdomen or intestinal obstruction.[1,2] The ileum is the most commonly reported site for duplication cysts accounting for over 60% of cases.[1,2]

Duplications of the colon are less common, accounting for 4–18% of all gastrointestinal duplications.[2] Alimentary tract duplications are considered to be benign lesions, usually asymptomatic, but may result in significant morbidity and mortality if left untreated. Complications such as bowel obstruction, gastrointestinal haemorrhage and malignant degeneration may occur. They may also be a diagnostic challenge especially when they present with complications such as haemorrhage or volvulus. Less than 100 cases of colonic duplications have been reported in the literature[3,4] and this is the first case from West Africa that the authors are aware of in a child with a colonic duplication.

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CASE REPORT

A 10-year-old girl presented at the children emergency with 3-day history of abdominal pain and vomiting. The pain was initially peri-umbilical but later radiated to the right iliac fossa. There was associated non-bilious, non-projectile vomiting and constipation. There was no history of fever.

Examination revealed a young girl, of asthenic build, well hydrated, in obvious painful distress. She weighed 25 kg and her temperature on admission was 37.6°C. She was tachycardic and tachypneic. There was moderate abdominal distention with tenderness in the right iliac fossa. Bowel sounds were reduced. Preoperative abdominal x-ray revealed multiple gas filled, grossly dilated bowel loops along the periphery of the abdomen. There was an associated S-shaped scoliosis of the vertebral spine. There were mottled luscencies along the right flank due to heavy faecal loading. Preoperative abdominal ultrasound scan showed significant gas reverberation in the right flank of the abdomen suggesting a segmental dilated bowel loop (ascending or transverse colon).

She was adequately resuscitated and had exploratory laparotomy done which showed a duplication cyst of the ascending colon – 25 cm in length twisted 180° anticlockwise around its mesentery with the caecum and appendix found in the left upper quadrant. She had



Figure 1: Dilated and twisted large bowel on plain x-ray



Figure 2: Duplicated colon segment in excised specimen

detorsion of the volvulus segment and resection of the duplication cyst along with the ascending colon and 10 cm of the terminal ileum. Single layer extra-mucosal ileo-colic anastomosis was affected with vicryl 3/0. Child did well post-operatively; she was commenced on oral feeds on 5th post operative day and was discharged on the 8th post operative day.

DISCUSSION

Alimentary tract duplications are congenital malformations that occur mostly in paediatric patients. The first reported case was made by Calder in 1733.[1] They have been variously referred to as enterogenous cysts, giant diverticula, ileal or jejunal duplex and the unusual meckel's diverticula.[2] In 1937, William E Ladd coined the term 'Duplications of the Alimentary Tract'.[5] Heiss[6] in a metanalysis of 12 large series encompassing 580 patients observed that 20% of these lesions occurred in the chest while 80% occurred in the abdomen, a small percentage were thoraco-abdominal. There are various classification systems – on the basis of origin; duplications may be described as foregut, midgut or hindgut derived depending on their locations.

Duplications of the colon constitute about 13% of all enteric duplications.[6] These lesions may be cystic (75%) or tubular (25%) in appearance and arise from the mesenteric border of the bowel.[3] They can be localised or extensive involving the entire length of the colon and emerging in the perineum as a separate opening sometimes in the back wall of the vagina. They may also be associated with other anomalies-conjoined twins, anomalies of the urinary tract, genitalia and lower spinal malformations.[2] Kottra and Dodds[7] described a classification system of hindgut duplications into short (type I) and tubular (Type II) duplications. The child in the index report had a tubular variety extending up to the transverse colon.

Histologically, most duplication cysts are lined by the mucosa native to the lesion, but ectopic tissue is present in 25–30% of specimens.[1] The most common type of ectopic tissue is gastric followed by exocrine and endocrine pancreatic tissue. Peptic ulceration causing perforation or haemorrhage may occur in duplications with ectopic gastric mucosa.[1] In children, duplications are considered as benign lesions but malignancy has been reported in adults.[1,3]

Duplications arise from disturbances in the embryonic development of the gastro-intestinal tract. Several major theories have been proposed for the formation of duplications at various sites, including the aberrant luminal recanalisation theory and the diverticular theory.[8] Others include the split notochord theory which explains the formation of neuroenteric duplications and associated vertebral anomalies. Recently, Koop[9] suggested that environmental factors such as trauma or hypoxia in early foetal development were likely to be responsible when multiple duplications are found in association with anomalies such as malrotation or atresia.

Presentation of colonic duplications is variable depending on size, shape and type of mucosa.[1] They may be asymptomatic and discovered accidentally at surgery. They may be minimally symptomatic and associated with vague abdominal pain, constipation or failure to thrive. They may present, as in the index case, as an emergency with acute intestinal obstruction due to sudden distention or volvulus-when there is an associated malrotation of the gut. Rectal bleeding is another emergency presentation when ectopic gastric tissue in the epithelial lining bleeds. While majority (80%) of patients present before 2 years of

age, duplication cysts may present at any age from the foetus to the geriatric patient.[10,11]

The diagnosis of colonic duplications may be difficult and the condition may be mistaken for acute appendicitis or intussuception. Plain abdominal X-ray as in the index case is non-specific and usually shows features of intestinal obstruction. Abdominal ultrasound scan may identify the cyst but is operator dependent. It demonstrates an inner hyperechoic rim of mucosal-submucosal tissues and an outer hypoechoic muscular layer.[12] Computerised tomography scan can help to localise the cyst and its relations with contiguous organs. An enhancing rim of tissue surrounding a fluid filled cyst is diagnostic; it may also help to identify synchronous lesions, if present. Barium enema may help in demonstrating communication with alimentary tract but may not be readily available in resource-poor setting for acute presentations.

Surgical treatment is advocated for all symptomatic duplication cysts.[1,13] Asymptomatic cysts discovered accidentally should also be resected to prevent future complications. Where possible as in the index case, complete excision of the cyst and the adjoining intestine should be carried out because of the intimate attachment of the common wall and because isolated resection of the cyst would compromise blood flow to the adjacent intestinal segment. In extensive tubular duplication cysts where excision may necessitate near total colectomy in a child, an alternative approach to management may be considered involving cyst marsupialisation, partial cystectomy and mucosal stripping[2]

The outcome after removal of most intestinal duplications is favourable. When bowel loss is kept to a minimum the effect on absorptive capacity is negligible.[13] Complications that may follow surgical resection are non-specific and include post-operative bleeding, infection and adhesive intestinal obstruction. Extensive resections may lead to chronic diarrhoea and malnutrition. However with proper treatment children born with enteric duplications do well and have excellent long-term outcomes and quality of life.[2]

Duplications cysts can be a diagnostic dilemma to the surgeon because of its varied presentations, a high index of suspicion is necessary to facilitate diagnosis, prompt operative intervention after adequate resuscitation may be necessary in a setting where diagnostic tests are either not available or affordable.

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