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ABSTRACT

Background: Gastrointestinal duplications are rare congenital malformations that commonly present within the first year of life. When they present in older age groups, diagnosis is often difficult. This difficulty derives from the lack of specific clinical features attributable to these lesions

Objectives: To document the challenges posed to surgeons by gastrointestinal duplications in different parts of the world and to report our experience in its management in Ile-Ife, Nigeria.

Design: A retrospective study. It involved a study of the clinical records of all patients diagnosed to have gastrointestinal duplication in our hospital.

Setting: A Paediatric and General Surgical unit, University of Ile-Ife Teaching Hospital, Ile-Ife, Nigeria..

Results: Five patients were diagnosed to have gastrointestinal duplications over the period of the study. The age, ranged from 28 days to 52 years. There were four males and one female. One patient presented within the first month of life. The others presented after the age of one year. There were two gastric, two ileal and one rectal duplication. All had the cystic variety of duplication. Three of the patients had excision, one had mucosal stripping and one had internal drainage of the duplication. The outcome was satisfactory in all of them.

Conclusion: Duplications tend to present at a much older-age group in our environment. Early diagnosis therefore depends on a high index of suspicion and every surgeon operating in the abdomen must be familiar with the management of this condition.

INTRODUCTION

Gastrointestinal duplications are rare congenital anomalies. They can arise anywhere in the gastrointestinal tract from the mouth to the anus(1). Presentation is usually within the first year of life. However some cases may present at an older age(2-4). Failure to recognise this condition in older patients may lead to missed diagnosis with serious consequences(5).

In spite of the rarity of this condition, many cases have been reported in the literature(5,6). However, most of these reports are from the developed world where the availability of modern investigative facilities has made preoperative diagnosis relatively easy. This in turn has allowed for planned surgical intervention in the management of affected patients. The aim of this study is to report the pattern of the clinical manifestations of gastrointestinal duplications and present our experience in the management of the condition in a tertiary health care facility in Nigeria.

MATERIALS AND METHODS

This was a retrospective study of all patients who had surgical treatment for gastrointestinal duplications in our hospital

between 1983 and 1996. Clinical records of all patients who had abdominal surgery during this period were reviewed and those with gastrointestinal duplications were further studied. Data pertaining to their age, sex, presenting symptoms and signs, methods of investigation, types of definitive treatment and outcome were analysed. All patients had exploratory laparotomy and diagnosis was confirmed by histology of excised specimens.

RESULTS

These are as summarised in Table 1. One adult and four paediatric patients had duplications. The paediatric cases constituted about one in 5,500 paediatric admissions. One patient with gastric duplication had excision of the septum and wide anastomosis between the cyst and the stomach. The other gastric duplication had excision and reconstitution of the stomach. One of the two patients with ileal duplication had excision of the duplication cyst and adjacent normal bowel, the other had right hemicolectomy due to the closeness of the cyst to the ileo-caecal junction and the involvement of the adjacent bowel in the inflammatory process occasioned by cyst rupture and peritonitis. The patient with rectal duplication was a 52 year old driver with a history of recurrent constipation from birth. He presented acutely with discomfort in the

Table 1

Characteristics of five cases of gastrointestinal duplications in Ile-Ife, Nigeria

No.	Presentation Age/Sex	Sign and symptoms	Pre-op. Diagnosis	Pathology of intestinal Duplications		Operation done	Associated congenital abn.	Outcome
				Site and type of duplication	Epithelia lining			
1	4/52 male	Bilious vomiting; visible peristalsis abdominal distension tender RIF mass	Intussusception	2 ileal cysts 15cm apart	Small intestinal epithelium	Bowel resection and anastomosis	Mesenteric cyst	Satisfactory
2	1 year 4 months, Female	Painless epigastric mass, weight loss, huge cystic intra abdominal mass	Mesenteric cyst	(1) Cystic mass arising from the anterior wall of the stomach contained 1500 ml of clear fluid (2) mesenteric cyst	Gastric type mucosa	Excision and primary closure	Mesenteric cyst	Satisfactory
3.	9 years, male	Recurrent RIF pain and mass. Weight loss. USS finding cystic RIF mass	Perforated Appendix	Cystic duplication of the ileum with peritonitis	Intestinal mucosa not specified	Right hemicolectomy	-	Satisfactory
4.	13 years, Male	Recurrent colicky, epigastric pain and vomiting, progressive weight loss emaciation, visible peristalsis and succussion splash	Gastric outlet Obstruction Secondary to Peptic ulcer disease	Cystic gastric mass in the greater curvature containing 200 ml of ash coloured mucoid fluid	Gastric epithelium	Excision of the septum	Absence of the LT upper rectus abdominis	Satisfactory
5.	52 years, Male	Chronic constipation alternating with diarrhoea, weight loss abdominal distension and visible peristalsis. Fluctuant retrorectal mass	Ischiorectal abscess	Rectal cyst	Columnar epithelium	Mucosal stripping of the cyst wall	-	Satisfactory

perineal region. He had exploration of the left ischiorectal fossa based on an initial diagnosis of ischiorectal abscess. The procedure was however abandoned when a soft mass that was thought to be arising from the bowel was found. Laparotomy and sigmoid colostomy were then carried out because he was thought to be suffering from Hirschsprung's disease. When he was re-explored for colostomy closure because histology of the colonic tissue obtained at the laparotomy had excluded Hirschsprung's disease, the retrocaecal mass was found to be persistent. Further dissection revealed this to be a cystic rectal duplication. Because of adhesions to other pelvic structures arising from the previous operation it was not possible to excise the cyst. The cyst was then opened and drained and its mucosal lining stripped off. All patients had an uneventful post-operative course.

DISCUSSION

Gastrointestinal duplications can arise anywhere from the mouth to the anus(1). Grossly, duplications are classified as being either cystic or tubular lesions. Cystic duplications are more frequent constituting about 94% of all cases in the literature(7). The cysts are located in the mesenteric border of the intestine, sharing a common blood supply with the normal digestive track adjacent to it(8). Cystic duplications vary in size depending on the location and duration of symptoms. It usually does not communicate

with contiguous loops of bowel. Tubular duplications on the other hand vary in length and communicate with the bowel lumen, usually at the distal end.

Gross(9) and Ladd(10) laid down histological criteria for diagnosing bowel duplications and these include;- presence of two muscular layers (longitudinal and circular) mesenteric cells and mucosa lining. The mucosa lining may be similar to the adjacent normal bowel or heterotopic. The mucosa in all the five cases reported in this series are similar to that of the contiguous bowel.

Clinical features vary depending on the anatomic location of the duplications and associated anomalies and complications. However, most cases reported in the literature present within the first year of life(8). Oesophageal duplications may present with cervical or mediastinal masses, cough, recurrent chest infections, respiratory difficulties and spinal anomalies such as anterior or posterior spina bifida.

Gastric duplications may present with epigastric tumour or gastric outlet obstruction(6) whereas intestinal obstruction, abdominal masses or peritonitis are the commonest presentations in small and large bowel duplications. Peritonitis may be as a result of perforation of the cyst wall. It may also be due to gangrene of the adjacent bowel due to obstruction of segmental blood supply(11). One of our patients presented with peritonitis arising from perforation of the cyst wall. Accumulation of secretion may give rise to tension and pain in the cyst.

Heterotropic gastric mucosa with communication of the cyst with the bowel lumen has also been reported to present with massive gastrointestinal bleeding.

The age at presentation of the five patients in this series varied from 28 days to 52 years with four presenting after one year of age. Male/female ratio was 4:1. This is at variance with reports in the literature where most patients present within one year of life with no sex predilection(12). However, Vanneuville *et al*(8) in their report of 13 cases found female; male ration of 9:4. All the five cases in this series were cystic. This is in keeping with findings in the literature where most cases are reported to be cystic(7). Various forms of associated malformations have been reported in patients with gastrointestinal duplications. These include ventricular septal defect, imperforate anus and malrotation of intestine(13). Two of our patients had associated mesenteric cyst and one had absence of the left rectus abdominis muscle(6).

Because of the protean manifestations, a high index of suspicion is needed in diagnosing gastrointestinal duplication. Diagnosis can be especially difficult when the patient is presenting for the first time in adulthood(14). The diagnosis was missed in our patient with rectal duplication presenting at the age of 52 years. Such missed diagnosis often results in wrong and multiple operations that increase the cost of health care and may prove disastrous for the patient.

Our experience shows that every surgeon should be familiar with this condition and be knowledgeable in the modalities of treatment. Cystic duplications are best excised along with the adjacent bowel. Tubular duplications that are not likely to lead to significant loss of bowel should be excised. However, long tubular duplications may pose a challenge in management. Such duplications are treated by stripping off the mucosa lining. This will remove long term complications of peptic ulceration, perforations, haemorrhage(15) and malignant transformation(1).

Three of our patients had excision of the duplication, one with rectal duplication had mucosa stripping and one

with gastric duplication was treated by internal drainage. Internal drainage is an acceptable form of treatment(16) for gastric duplication but the preference should be for an operation that completely removes all the cyst lining because of the risk of malignant transformation(1).

REFERENCES

1. Johnson J.A. 3rd, Poole G.V. Ileal duplications in adults: *Arch. Surg.* 1994; **129**:659-661.
2. Glaser C., Kuzinkovas V. and Maurer C. *et al.* A large duplication cyst of the stomach in an adult presenting as pancreatic pseudocyst; *J. Gastroent. Hepatol.* 1998; **15**:703-6.
3. Estlain D. and Wolloch Y. Gastric duplication cyst in an adult. *Harefuah* 1997; **132**:634-635.
4. Frering V., Vealecala E. and Fougue P. *et al* Upper digestive duplications in adult. *Ann. Chir.* 1995; **49**:925-935.
5. Oluwole S.F. and Adekunle A. Rectal duplication in chronic large bowel Obstruction. *Can J. Surg.* 1987; **30**:419-420.
6. Adejuyigbe O., Hamed A.O. and Fadiran O.A. Gastric duplication: case report and review of the literature. *Nig. Med J.* 1988; **18**:357-361.
7. Mcletchie G.B., Purris J.K. and Saunders R.C. The genesis of gastric and certain intestinal diverticula and enterogenous cyst. *Surg. Gynec. Obstet.* 1954; **99**:135.
8. Vanneuville G., Scheye T. and Dechelotte P. *et al.* Duplications of the alimentary tract in children. *Chirurgie Int.* 1996; 15-18.
9. Gross R.E. Duplications of the alimentary tract in the surgery of infancy and childhood; Saunders Philadelphia PA, pp. 221-245, 1953.
10. Ladd W.E. Duplications of the alimentary tract. *South Med. J.* 1937; **30**:363-371.
11. Yang M.C., Duh Y.C. and Lai H.S. *et al.* Alimentary tract duplications. *J. Formos. Med. Ass.* 1996; **95**:406-409.
12. Pinter A.B., Schubert W. and Szemledy F. *et al.* Alimentary tract duplications in infants and children. *Eur. J. Paediat. Surg.* 1992; **2**: 8-12.
13. Iyer C.P. and Mahour G.H. Duplications of the alimentary tract in infants and children. *J. Paediat. Surg.* 1995; **30**:1267-1270.
14. Ildstad S.T., Tollerud D.J. and Weiss R.G. *et al* Duplications of the alimentary tract clinical characteristics, preferred treatment and associated malformations. *Ann. Surg.* 1988; **208**:184-189.
15. Nagar H. Duplications of the gastrointestinal tract. *Isr. Med. Ass. J.* 1999; **1**:254-6.
16. Taft D.A. and Hairston J.T. Duplication of the alimentary canal. *Ann. Surg.* 1978; **42**: 455-4 62.