Case Report

PYLORIC DUPLICATION CYST: A RARE CONGENITAL ANOMALY

Altaf Hussain Rathore[1], Riaz Hussain[2], Farhan Rathore[3]

ABSTRACT

Pyloric duplication cyst is one of the rarest congenital anomalies. We present such a case in a boy of 6-1/2 years old who was treated by us successfully. The theories of its embryogenesis, mode of presentations, diagnosis and treatment are discussed.

KEY WORDS: Duplication of stomach, Pyloric duplication cyst.

Pak J Med Sci April - June 2010 Vol. 26 No. 2 494-496

How to cite this article:

Rathore AH, Hussain R, Rathore F. Pyloric duplication cyst: A rare congenital anomaly. Pak J Med Sci 2010;26(2): 494-496.

INTRODUCTION

Duplication of stomach is a rare congenital anomaly and a very few cases of pyloric duplication cyst have been reported.[1] It varies in size and location but in majority of the cases it is located at the greater curvature.[2] It shares a common wall with normal stomach and may or may not communicate with the stomach. When it does not communicate with any structure it is called gastric duplication cyst; if adjacent to pylorus it may also be called pylorus duplication cyst like our case. Majority of these cysts are ovoid or spherical. Three theories have been presented for its embryogenesis;

1. Prof. Dr. Altaf Hussain Rathore, FRCS, Foundation Hospital Rajana
2. Prof. Riaz Hussain, FRCS, Punjab Medical College Faisalabad
3. Dr. Farhan Rathore, MD, Foundation Hospital Rajana, District Toba Tek Singh, Punjab.

Correspondence

Prof. Altaf Hussain Rathore
E mail: foundationhospital@hotmail.com

* Received for Publication: August 8, 2009
* Revision Received: January 5, 2010
* 2nd Revision Received: January 15, 2010
* Final Revision Accepted: March 15, 2010

A. *Bremers Theory*[3] :Embryological persistence of vacuoles within foregut wall.
B. *Lewis and Thyng*[4] :Due to persistent embryogenesis diverticulum seen in lesser curvature.
C. *Mcletchie Theory*[5] :It is due to faulty s eparation of endoderm and notochord in early development.

It may be communicated with pancreatic duct[6] so may cause relapsing pancreatitis.[7] It may give rise to serious complications like ulceration perforation, hemorrhage, gastric outlet obstruction, malignancy and pseudopancreatic cyst.[8] The usual symptoms are pain epigastrium, vomiting, vague mass upper abdomen, weight loss, failure to thrive and malaena.[9] Surgery is an undisputed mode of management and so many modification have been offered by different authors.[10]

CASE REPORT

A six years old boy presented to us on 28-04-09 with pain upper abdomen, lump and vomiting off and on since birth. He could take only milk and fluids and he used to feel relief in pain after vomiting. So he failed to gain height and weight. He used to have constipation some times but his urination was normal. The parent

consulted so many doctors and pediatricians of the teaching hospitals and numerous investigations were done including CT Scan but no body could pin point the disease. All gave him different medicines but none of them examined the patient properly. At the time of examination he had an ill defined lump just above right side of umbilicus.

Blood Report: Hb was 9G, ESR 30 per hour and blood group AB+.

Ultrasound Report: Increased mural thickening of transverse colon distal to hepatic flexure with thickness of 6mm of 40mm long segment. No separate visible soft tissue mass, no volvulus no intussusceptions, no free fluid in the peritoneal cavity.

CT Scan Abdomen: With oral contrast ill defined rounded mass with central low attenuation zone observed in right hypochondrial region inseparable from serosal region of anterolateral wall of transverse colon measuring 35mm x 33 mm in axial dimensions and 42mm in vertical dimensions, no relation to gastric wall, duodenum, gall bladder or liver observed. Pancreas was normal.

Operation: He was operated on 08-05-09 under general anaesthesia. A long right paramedian incision was given there was a thick wall cyst of size of a golf ball having common muscular layer of the pylorus inseparable from transverse colon. The character of mucosa and muscular layer of the cyst was more like stomach. It had no communication with the stomach and colon. The cyst along with part of stomach and 10cms of transverse colon was removed and primary repair of stomach and colon was performed. 200ml of blood was transfused.

Post Operative: Patient made an uneventful recovery. He was discharged on 9th day after operation after removal of sutures with perfect healing. He reported in the outdoor 7 days later. He started eating solid food and had a ferocious appetite and has started gaining the weight.

Biopsy Report:

Gross Appearance: (Fig-1). The specimen consists of a 10cm long segment of intestine. Close to it there is a cystic mass with central lumen. The lumen is not in continuity with that of intestine or stomach. The thickness of wall is 4-5mm. Two sections taken from the cystic wall.

Microscopic appearances: The section shows wall of stomach with all layers i.e. mucosa, muscularis mucosa brunner glands and muscularis externa. The mucosa has tall columnar epithelium. There is no inflammatory or any significant abnormality.

Opinion: Gastric Wall

DISCUSSION

Incidence of duplication of stomach is much rare and least expected. The failure to thrive and firm lump above umbilicus made us suspicious of this disease. Ultrasound and CT Scan has been the most useful investigation to diagnose this anomalies with other author[11,12] but both were misleading in our case. Non bilious vomiting is the main complaint in infants in this disease but is more common in pyloric stenosis which made the diagnosis difficult.[13] That is why the disease was diagnosed at laporotomy. Other more useful investigations in this disease like EUS (endoscopic US) and MRI[12] were not available to us. Most of the cases are diagnosed in infancy[14] but our case could not be diagnosed till the age of six year. Gastric duplication is

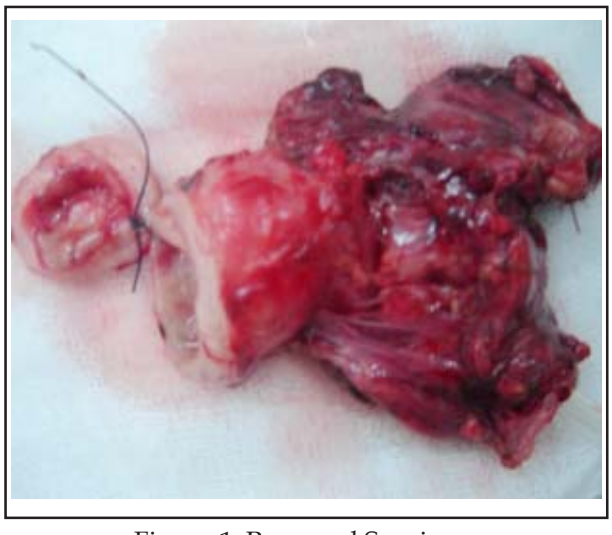

Figure-1: Removed Specimen

Note: Opened Cyst riding on the top of the excised regent of transverse colon.

more common in females[15] though our case was a male. It is reported to be associated with other anamolies in 50% of the cases especially of spine and gastrointestinal tract[16] but there was no other congenital malformation in our case. We removed the cyst in toto as recommended by most of the authors.[17] We had to remove the part of pylorus and a small segment of transverse colon as both could not be separated from the cyst. Primary repair of colon and stomach was done. As pressure on both visera was relieved so he started taking normal food without any symptoms.

CONCLUSION

This was a rare case of congenital pyloric duplication cyst which was partially blocking stomach and transverse colon but had no communication with anyone of them. His all symptoms were due to pressure which got relieved after its removal.

REFERENCES

1. Tang XB, Bai YZ, Wang WL. An intraluminal pyloric duplication cyst in an infant. J Pediatr Surg 2008;43(12):2305-7.
2. Cooper S, Carbaugh RA, Abram AS, Pyloric duplication: Review and Study. Am Surg 1995;61:1092-1094.
3. Bermer JL. Congenital anomalies of the viscera. Cambridge, MA: Harvard University press 1957.
4. Lewis FT, Thyng FW. Regular occurrence of intestinal diverticula in embryos of the pig rabbit. Am J Anat 1907;7:505.
5. McLetchie NGB., Rurves JK, Saunders RL.de C.H Genesis of gastric and certain intestinal diverticula and enterogenous cysts. Surg Gynecol Obstet 1954;99:135-141.
6. Hishiki T, Saito T, Terui K, Mitsunaga T, Nakata M, Matsuura G, et al. A rare presentation in a case of gastric duplication cyst communicating to the pancreatic duct: coincidental detection during pyloromyotomy for hypertrophic pyloric stenosis. J Pediatr Surg 2008;43(9):e1-3.
7. Lavine JE, Harrison M, Heyman MB. Gastroenterol duplication causing relapsing acute pancreatitis in children. Gastroenterol 1989;97:1556-1558.
8. Kremer RM, Lepott RB, Izant RJ Jr. Duplication of the stomach. J Pediatr Surg 1970;5:360-364.
9. Parker BC, Guthrie J, France NE, Attwell JD. Gastric duplication of the alimentary tract. Br J Surg 1981; 68:92-96.
10. Iwasaki M, Nishimura A, Kamimura R, Ura K, Kobayashi H, Saiga T. Pyloric duplication cyst in an infant. Pediatr Int 2009;51(1):146-9.
11. Gupta AK, Beny M, Mitra DK: Gastric duplication cyst in children report of 2 cases. Pediat Radiol 1994;24;346-347.
12. Takahara R, Torigoe T, Haga H, Gastric duplication cyst, evaluation by endoscopic Ultrasonograpy (EUS) and Magnetic Resonance imagining (MRI). J Gastroenterol 1996,31;420-424.
13. Upadhyaya VD, Srivastava PK, Jaiman R, Gangopadhyay AN, Gupta DK, Sharma SP. Duplication cyst of pyloroduodenal canal: a rare cause of neonatal gastric outlet obstruction: A Case Report. Cases J 2009;2(1):42.
14. Moss RL, Hatech EL, Kozarek RA, Riyan JA. Panceratitis caused by gastric duplication communicating with an aberrant pancreatic lobe. J Pediatr Surg 1991;31:733-736.
15. Pruksopon C, Donovan RT, Pinit A. Gastric duplication. J Pediatr Surg 1979;14:83-85.
16. Dudgeon DL. Lesions of the stomach in Ashcraft, Holder (Eds) Pediatric Surgery, 2nd ed. Chap. 1993;24:295-297.
17. Ravitch M. Duplication of the gastrointestinal tract in Welch KJ et al. Pediatri Surg, 4th ed. 1986,914-915.