LEIOMYOSARCOMA ARISING IN DUPLICATION CYST OF SMALL BOWEL PRESENTING WITH LOWER URINARY TRACT SYMPTOMS. A CASE REPORT AND LITERATURE REVIEW

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INTRODUCTION

Bowel duplication is a rare congenital anomaly which may involve the gut anywhere from mouth to anus but small bowel is the most common site affected. The disease usually presents in infants and children but may present in adults as a consequence of complications. Extra abdominal enterogenous cysts have been found, such as in CNS, retroperitoneum, mediastinum and pancreas which create significant difficulties in the diagnosis and management at such unusual situations.[1] Many complications can be encountered in the cyst, such as haemorrhage, torsion,, volvulus of bowel, intestinal obstruction, mass effect on other organs, and malignant changes.[1-4] A case of duplication cyst of small bowel with malignancy (leiomyosarcoma) is reported in a 55 years old man who presented with the clinical features of enlarged prostate.

CASE REPORT

A 55 years old man resident of Afghanistan was admitted to Surgical "C" unit with 4 months history of urinary symptoms of frequency and dysuria and was diagnosed as a case of bladder neck obstruction due to enlarged prostate.

1. Examination of this elderly man, physically fit, revealed a globular cystic mass in lower abdomen, slightly mobile from side to side, arising from pelvis.

The mass was 15 x15 cm, partly dull and partly resonant to percussion. On rectal examination a firm enlarged prostate was palpable and a mass could be felt at the tip of finger above and behind the bladder.

Laboratory tests were as follow:- Haemoglobin 11gm%, TLC 8500/cm. Blood urea 100gm%. Blood sugar 60mg%, urine examination revealed pus cells (9-6/HPF) blood cell occasional. X-Ray chest, KUB, ECG were normal.

2. On exploration of lower abdomen a large cystic mass was found intraperitonially wedged in pelvis behind urinary bladder and in front of rectum.

Urinary bladder was found empty and compressed. The mass was found to be cystic duplication of gut, attached to mid ileum and eroding posterior wall of urinary bladder. Whole of the cystic mass along with the attached small bowel was resected and end to end small bowel anastomosis established. The eroded region of the posterior wall of the bladder was also resected and closed in two layers.

Urinary bladder was drained with urethral catheter. Abdomen was closed with a drain in pelvis. Postoperatively patient developed wound sepsis which was cleared by local and systemic treatment. Resected mass was opended and found to contain masses of grey whitish friable tissue and diarty foul smelling greenish black fluid with food particls. Histopathology revealed leiomyosarcoma in the wall of the duplication cyst.

DISCUSSION

Bowel duplication or duplication cyst of bowel (enterogenous cyst) has a variety of presentation at different ages of life. In a series of paediatric patients, 80% presents in the neonatal period.[5] A number of theories have been proposed to the cause of this rare congenital abnormality including diverticular theory, sequestration of primitive gut, error in solid cord stage of bowel development and abnormal growth of the notochord.[6]

Small bowel duplication is commoner than the rest of the bowel. Extra abdominal duplication cysts are rarely found in the mediastinum, retroperitioneum and central nervous system which result in great difficulties in the diagnosis and management of these patients.[1,7]

Manifestation of the bowel duplication is dependant on its size and location. Many abdominal duplications produce palpable masses. Most of the symptoms are produced by torsion or volvulus of the bowel, compression of the bowel, perforation and haemorrhage into the bowel, rupture of the cyst, mass effect obstructing and displacing other organs of the body such as rectum, bladder and uterus.[8] Malignancies such as adenocarcinoma and squamous cell carcinoma have been reported in the wall of duplication cyst.[1,2,3] Ultrasound and CT Scan has frequently provided detailed information compared to other conventional contrast studies. Technetium scan can be used to show ectopic gastric mucosa lining the duplication cyst which can be of significant help in the diagnosis of these cysts in unusual sites such as retroperitoneum.

Once diagnosed, these cyst can be excised totally alongwith part of bowel with end to end anastomosis, stripping of the mucosal from tubular duplication or anastomosis of the distal end of the tubular duplication with the intact bowel averting the possible risk of malignant changes.[8]

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