Case Snippets

Achalasia cardia In mother and son

Atul Sachdev, Elmajlit S Sangdu, Sanjay D’Cruz, Sarbmeet S Lehl, Vikas Agarwal

Department of Medicine, Government Medical College and Hospital, Sector 32-B, Chandigarh 160 030

Familial occurrence of achalasia cardia is rare. Most associations are among siblings or in monozygotic twins. Parent-child association is even rarer and only six such instances have been reported till date. We report a 29-year-old man with achalasia cardia and his mother who had the same illness two and half years later. Both of them were successfully treated with balloon dilatation. [Indian J Gastroenterol2004;23:109]

Key words: Esophageal motility disorder, familial predisposition or a common environmental factor. Most of the familial associations reported are among siblings or in monozygotic twins, thereby suggesting horizontal transmission of the disease. This is supported by the occurrence of disease in consanguineous families at an early age. The mode of inheritance suggested is possibly autosomal recessive, but is however unproved.

The occurrence of vertical transmission is also rare. A literature search has revealed only six such instances. This may suggest either an autoimmune disease, which has manifested in parent and child, or a common environmental agent, which triggered the disease in both.

In conclusion, this report documents a rare occurrence of achalasia cardia in both parent and child.

References


Correspondence to: Dr. Sachdev, Professor and Head, Department of Medicine, #1104, Medical College Campus, Sector 32-B, Chandigarh 160 030. Fax: (172) 266 8468. E-mail: atulasachdev@glide.net.in

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Bouwer's syndrome – an unusual cause of spontaneous resolution of gastric outlet obstruction

Gajanan D Wagholkar, Mohammad Ibrarullah

Department of Gastroenterology, Sri Venkateshwar Institute of Medical Sciences, Tirupati 517 507

Gastric outlet obstruction due to a gallstone impacted in the duodenal bulb (Bouwer's syndrome) is a rare complication of gallstones. We report a 47-year-old man with this syndrome in whom the impacted stone migrated uneventfully. [Indian J Gastroenterol 2004;23:109-110]
Key words: Cholecysto-enteric fistula, gallstone il- eus

Bouveret's syndrome (BS) is a rare complication of gallstones, characterized by gastric outlet obstruction due to a large stone impacted in the proximal duodenum consequent to a cholecysto-duodenal fistula.1,2

A 47-year-old man presented with epigastric pain, recurrent vomiting and low-grade fever since three days. He was diagnosed to have cholelithiasis on ultrasonography performed six months earlier for dyspeptic symptoms. Examination revealed mild dehydration. Vital signs were within normal limits. There was minimal tenderness in the epigastrium and right upper quadrant. Hematological and biochemical investigations including liver function tests were normal. Esophagogastroduodenoscopy revealed an extrinsic bulge in the antro-pyloric region. Further manoeuvring of the endoscope showed a large yellow-brown, oval stone impacted in the duodenal bulb and causing obstruction (Fig. 1a). An attempt to disimpact the stone with foreign body forceps failed. Dormia basket retrieval too failed since it could not be passed beyond the impacted stone. The patient was managed conservatively with naso-gastric aspiration, intravenous fluids, and broad-spectrum antibiotics.

Repeat endoscopy the day after showed pus in the duodenal bulb. There was, however, no evidence of the stone (Fig 1b). The duodenal mucosa was inflamed. The endoscope could be easily negotiated into the second and third parts of the duodenum. Ultrasonography revealed a thick-walled gall bladder with minimal pericholecystic fluid collection but no stone in the gall bladder. Plain X-ray of the abdomen did not reveal any ectopic radio-opaque gallstone. The symptoms resolved dramatically. He continues to be symptom-free two years later with no further stones in the gall bladder.

In a majority of cases gallstones that enter the intestinal tract through a cholecysto-enteric fistula are passed spontaneously; 6% develop clinical obstruction. The terminal ileum is the commonest site (60%), followed by the proximal ileum (24%) and jejunum (9%). In 1%-3% of cases the stone may obstruct the duodenum, usually in its distal portion. Impaction in the pyloric region and duodenal bulb causing gastric outlet obstruction (Bouveret's syndrome) is the least common.

The classical triad of distended stomach, pneumobilia and ectopic radio-opaque gallstone on plain X-ray of the abdomen is diagnostic of Bouveret's syndrome but is seen in only a third of cases.3 Pneumobilia may not always be detected and gallstones are radiolucent in the majority of cases.1 Contrast radiology, CT scanning or upper GI scopy are required to confirm the diagnosis.

Endoscopy is rarely therapeutic, because it is difficult to dislodge and retrieve the large impacted stone. Recently successful treatment using laser lithotripsy4 has been reported. Surgical removal of the offending stone is the most accepted treatment.1,2 Cholecystectomy and repair of fistula in addition to enterolithotomy is recommended. This is debatable as the morbidity related to the bilio-enteric fistula is usually low.2 The present case was unique in that the patient had a fortuitous and uneventful passage of the impacted stone, which to our knowledge is the first report in literature of spontaneous resolution of Bouveret's syndrome.

References

Correspondence to: Dr Waghjilkar, Assistant Professor, Department of Gastroenterology. Fax: (877) 228 6803. E-mail: drgajjan2002@yahoo.com
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Perforation of jejunal non-Hodgkin's lymphoma

Minakshi Ishwar Jambhulkar,
Mohan Achyut Joshi, Dharmesh Balsarkar,
Mahendra Chandak, Sanjay Pareb

Department of Surgery, Lokmanyaa Tilak Municipal Medical College, Mumbai 400 022

Primary gastrointestinal lymphomas are rare. Jejunal non-Hodgkin's lymphoma presenting as perforative

Fig 1: (a) Large gallstone impacted in duodenal bulb causing obstruction; (b) repeat evaluation revealed pus in duodenal bulb with inflamed mucosa; the stone had migrated spontaneously

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