Brunneroma presenting with radiological features of duodeno-duodenal intussusception


1 Registrar in Surgery, Colombo South Teaching Hospital, Kalubowila, Sri Lanka.
2 Senior Registrar in Surgery, Colombo South Teaching Hospital, Kalubowila, Sri Lanka
3 Consultant Surgeon, Colombo South Teaching Hospital, Kalubowila, Sri Lanka

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Introduction

Benign tumours of the duodenum are exceedingly rare (incidence <0.01). Approximately 11% of these are Brunner's gland adenoma/Brunneroma or polypoidal hamartoma which is a benign, proliferative lesion of the submucosal Brunner's glands [1]. Duodeno-duodenal intussusception (DDI) is a rare presentation of such a tumour with only a few cases reported thus far. We present a case of a large Brunneroma in the first part of the duodenum (D1), with radiological features of DDI. However the clinical symptom profile and intra-operative findings were not suggestive of acute or chronic duodenal obstruction.

Case presentation

A 67 year old lady presented with a history of melaena of 1 week duration with a vague, intermittent epigastric pain with regurgitation for more than a year. Examination was unremarkable except for the marked pallor. Iron deficiency anaemia was confirmed with haemoglobin 6.6g/dl and serum ferritin 21 μg/L.

Abdominal ultrasonography revealed a 4.3 cm × 3.2 cm × 4.4 cm hyperechoic mass in left hypochondrial region and CECT of the abdomen suggested a short segment of duodenum appeared intussuscepted into the proximal portion of the third part of duodenum (D3) (Figure 1). The distal part of D3 appeared intussuscepted into the D4 segment. No definite lead point demonstrated and no significant proximal or distal bowel distension seen to suggest obstruction.

In Upper gastrointestinal endoscopy a mobile polypoidal mass was identified on the postero-superior wall of D1 (Figure 2). Histology of the endoscopic punch biopsy was inconclusive. Polypectomy was performed through a gastrotomy incision by mobilizing the pedunculated polyp (Figure 3) in to the stomach from the posterior wall of D1. Histopathology confirmed a Brunneroma with a diameter of 6 cm. Neither during endoscopy nor surgery was there evidence of DDI.

Discussion

Duodeno-duodenal or duodeno-jejunal intussusception is an extremely rare manifestation with only a handful of reported cases [2]. Entero-enteric intussusception itself is a rare event in adults and this is always secondary to the presence of a lead point like a hamartoma, Meckels diverticulum or a tumour. Cruveilhier described one of the earliest cases of Brunner gland adenomas co-incidentally associated with fatal duodenal intussusception [3].

DDI is exceptionally rare because the retroperitoneal situation fixes the duodenal wall. The radiological diagnosis of duodenal intussusception is based on characteristic features seen in contrast upper GI series, CT and ultrasound. The classic 'coiled spring sign' usually apparent in contrast films of jejunal intussusception, is not so easily evident in the duodenum. CT or ultrasound evidence of duodenal intussusception is based on the appearance of the 'target sign' due to the multi-layered appearance of the telescoping bowel walls [4].
In our case the CT was suggestive of DDI in the proximal and distal parts of the duodenum, but she had no features to suggest either acute or chronic duodenal obstruction. Despite the retroperitoneal fixation, as a result of mucosal elongation or slipping, duodenal tumours can migrate even up to the jejunum [5]. Saida et al while reviewing a series of duodenal tumours makes the interesting submission that some cases of duodenal intussusception based radiological signs may not be due to a true intussusception but a radiological illusion due to mucosal prolapse as a result of mucosal elongation or slipping [6]. It is possible that this was the case in our patient as well. Alternatively it may be that there was spontaneous resolution of a true duodenal intussusception.

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References

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**Key Learning Points**

- Adult intussusception including duodeno-duodenal intussusception is a rare event which is always secondary to the presence of a lead point like a tumor or diverticulum.
- Duodenal intussusception based radiological signs may not be due to a true intussusception but a radiological illusion due to mucosal prolapse.