CASE REPORTS

Intraluminal duodenal diverticulum causing recurrent pancreatitis: treatment by endoscopic incision

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Abstract
Intraluminal duodenal diverticulum is a recognised but rare cause of acute pancreatitis. This patient had three attacks of pancreatitis, each requiring a stay in hospital, within a four month period. The apex of the diverticulum was incised endoscopically, whereupon peas and food debris gushed from the incision site. The patient has had no further symptoms in the 12 months since the endoscopic procedure.

Intraluminal duodenal diverticulum is thought to be part of the spectrum of congenital abnormalities, which includes duodenal atresia. It usually presents in adults, after enlargement of the diverticulum with food or ingested foreign bodies, resulting in obstruction of the duodenal lumen, or more rarely of the ampulla of Vater. Definitive treatment has historically been surgical, but we report on a patient with recurrent pancreatitis in whom a satisfactory outcome was achieved with an endoscopic sphincterotome. As far as we are aware, this technique has not previously been reported for treatment of an intraluminal diverticulum.

Case report
A 41 year old man in previous good health was admitted with a seven hour history of central abdominal pain radiating to the back, and vomiting. He drank little alcohol and was receiving no treatment. Clinical examination showed tenderness in the epigastrium but no other abnormality, but investigations confirmed a diagnosis of acute pancreatitis (amylase 2830 U/l (normal <200)). Liver enzymes were mildly deranged: γ-glutamyltransferase 123 U/l (<65), alanine amino transferase 183 U/l (<45), alkaline phosphatase 92 U/l (<130), bilirubin 29 µmol/l (<17). Upper abdominal ultrasound did not show any abnormality and the maximal bile duct diameter was 6 mm. The abdominal pain and vomiting settled quickly after intravenous fluids were given, and within four days the patient was discharged home. He remained well for two months and then required readmission with a further bout of pancreatitis (amylase= 2570 U/l), which settled with conservative management. Abdominal computed tomo-

graphy showed changes compatible with acute pancreatitis but no other abnormality and plasma calcium and lipid analyses were repeatedly normal. Endoscopic retrograde cholangiopancreatography (ERCP) was scheduled, but before this could be performed, he had a further attack of pancreatitis, which again settled quickly.

At ERCP a small (approx 7 mm diameter) diverticular opening was seen at the presumed site of the ampulla of Vater, with a 5 cm long polypoid swelling protruding into the duodenal lumen distal to the ampulla (Fig 1). The mucosa overlying this swelling looked normal, but was baccate (as if containing spherical objects). When prodded with a cannula the texture was soft and compressible. This was initially felt to be a prolapsing ampulla, possibly with a stone at the lower end of a dilated bile duct, although the possibility of an intraluminal diverticulum was considered. The ampullary opening could not be located, and so a small precut was carefully made at the apex of the swelling. On enlarging the opening with a conventional sphincterotome, several peas and other food debris gushed into

Figure 1: Photograph taken at endoscopy showing a polypoid intraluminal swelling whose texture is being assessed by the application of an endoscopic retrograde cholangiopancreatography cannula. A diverticular opening was seen at the proximal end of the swelling (arrow) but the ampulla of Vater was not identified. An opening was made at the distal end of the swelling, permitting the release of several peas and other debris.
the lumen of the duodenum. Neither a cholangiogram nor a pancreatogram were obtained as the ampulla was never located, presumably because it lay within the neck of the diverticulum. Review of a previous barium meal (Figs 2 and 3) showed the presence of an intraluminal diverticulum that had been overlooked and a subsequent video barium meal showed contrast entering a diverticulum and leaving it by the distal opening, which resulted from the endoscopic incision. The diverticulum was seen to contract peristaltically. The endoscopic and radiological findings are diagnostic of intraluminal duodenal diverticulum, and the patient has remained well for 12 months after the endoscopic procedure.

Discussion
The foregut lumen of the human embryo is occluded by epithelial cells, and normal development is characterised by recanalisation after seven weeks. Failure of the normal development in the duodenum can result in various congenital anomalies including duodenal atresia, stenosis, mucosal diaphragms, and duodenal duplication. Many such abnormalities present in childhood and are surgically treated. Whether intraluminal duodenal diverticula arise as a congenital abnormality in themselves, or result from gradual peristaltic pulsion acting on a duodenal diaphragm has been the stimulus for considerable debate. The evidence for the second hypothesis is that intraluminal duodenal diverticulum occur in adults, not children, and that the orifice of the diverticulum is always proximal with the fundus distal. Whether a lesion should correctly be termed a duplication or diverticulum remains a vexed question. Bremer differentiated congenital duplications from diverticula by the presence (in the first) of all the normal intestinal layers for the greater part the wall, whereas Wiot and Spiro concluded that the important differentiating factor was that diverticula were in continuity with the main intestinal lumen. Associations with other congenital lesions including Down’s syndrome, intestinal malrotation and hernias, and vascular abnormalities have been reported in previous reports.

In contrast with the commoner outpouching diverticula, intraluminal ones are often symptomatic. Typical symptoms, in the absence of specific complications, are those of partial or intermittent duodenal obstruction, with postcibal fullness or pain relieved by vomiting. Reported complications include haemorrhage from ulceration within diverticula, and cholangitis or pancreatitis, which are thought to occur as a result of occlusion of the respective duct by an enlarging pouch.

Diagnosis may be suspected at endoscopy when the picture may be that of a blind sac, which when inverted seems like a polyp. The endoscopic appearance excludes a diagnosis of duodenal varices, and ampullary tumours would have the appearance of a mass lesion on barium studies, compared with the hollow sac found in our patient. Endoscopy alone may not always distinguish between extra and intraluminal diverticula and barium studies are usually definitive. In contrast, duodenal duplications do not have a prominent diverticular orifice, and the appearance of barium studies is that of a mass lesion, which does not collect food or barium (but may contain gall stones).
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Cholelithiasis has many similarities to a duodenal duplication in being periampullary in site, communicating with the ampulla and bilary tree, but differs by the fact that it is internally lined by biliary rather than duodenal epithelium. Without exciting the lesion or performing full thickness biopsies, it is not possible to make a diagnosis of intraluminal duodenal diverticulum unequivocally, but the presence at endoscopy of a diverticular orifice, peristalsis of the sac, and the collection within the sac of food and barium, suggest that our patient's abnormality is in keeping with this diagnosis.

Several modes of treatment for intraluminal diverticula have been proposed. Although these lesions are rarely asymptomatic, many of the symptoms relate to impaction of undigested food, and removal of the impacted food, together with strict instructions about mastication may suffice in some elderly patients. Endoscopic removal of a foreign body from within the diverticulum is satisfactory for a small number of cases and if bile duct calculi are present these should be removed but most patients will require a definitive procedure. Endoscopically, this can be achieved using a polypectomy snare as described by Hajiro et al. although extreme care must be taken to avoid snaring the papilla.

In our patient, the papilla could not be located so the approach of Hajiro et al would have been treacherous. It seemed logical to open the apex of the diverticular sac using a precut sphincterotomy knife, followed by extension using a conventional sphincterotomy wire, as described for treatment of duplication cysts. Surgical excision of the diverticulum has been advocated by some, but here again extreme caution should be taken, as a number of other local anatomical anomalies may be associated. In our view this is evidence to support an endoscopic approach to the treatment of the intraluminal diverticulum, although the ultimate method chosen may depend partly on available expertise.