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Case report

Pseudolipoma of inverted Meckel's diverticulum: clinical, radiological and pathological correlation

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Abstract. Three cases of isolated inverted Meckel's diverticulum are described. In two cases an initial pathological diagnosis of small bowel lipoma was suggested. In a third case central fat was demonstrated on CT and peristalsis of the intraluminal polypoid mass was observed during US examination. In all three cases small bowel enema examination demonstrated the lesion. Correlation of the clinical, radiological and pathological features is emphasised, as this will allow the correct diagnosis.

Key words: Inverted Meckel's diverticulum – Lipoma – Small bowel – Radiology – Ultrasonography

Introduction

Intussusception is a well-recognised complication of Meckel's diverticulum and in virtually all cases Meckel's diverticulum is found to be inverted [1, 2]. Isolated inversion of Meckel's diverticulum without intussusception is apparently exceedingly rare. We illustrate how this condition can simulate small bowel lipoma, and suggest that the incidence may be higher than currently considered. Correlation of the clinical, radiological and pathological findings in three cases allowed the correct diagnosis.

Case reports

Case 1

A 39-year-old man presented with a 2-month history of non-specific lower abdominal pain and a single episode of rectal bleeding. Physical examination revealed mild tenderness in the right iliac fossa. Routine haematology and biochemistry showed an iron-deficiency anaemia – haemoglobin (Hb) 10.5 g/dl – but other parameters were normal. Crohn's disease was suspected clinically and the patient was referred for small bowel enema (SBE). This showed a polypoid filling defect in the distal ileum (Fig. 1 a). At surgery an umbilicated lesion was present on the antimesenteric border of a distal ileal loop (Fig. 1 b). A section of small bowel was resected and opened to display a polypoid tumour (Fig. 1 c) consistent with an inverted Meckel's diverticulum. Consequently, the preliminary gross pathological impression of a lipoma was surprising (Fig. 1 d). However, histological examination revealed a subserosal location of the adipose tissue consistent with invaginated mesenteric fat (Fig. 2 b). In addition, the mucosa overlying the tumour was extensively ulcerated with foci of heterotopic gastric-type mucosa, small islands of pancreatic tissue and microscopic clusters of neuroendocrine cells representing a small carcinoid tumour. The final diagnosis was an isolated inverted Meckel's diverticulum.

Case 2

A 59-year-old man presented with a 4-month history of increasing shortness of breath on exertion and angina. There were no abdominal symptoms. He was noted to have a severe iron-deficiency anaemia (Hb 7.3 g/dl) and faecal occult blood testing was positive. Upper gastrointestinal endoscopy and barium enema were normal. An SBE revealed a polyp in the distal ileum (Fig. 3) which was resected. Histologically, the lesion was thought to be a subserosal lipoma. The patient was discharged from follow-up 6 months later with no further symptoms (Hb 16.0 g/dl). No mention of Meckel's diverticulum was recorded in the patient's notes; however, the operation details recorded "normal laparotomy apart from a polypoid lesion in ileum (2 feet from ileocecal valve)". The histology was reviewed and the subserosal location of the adipose tissue confirmed. The overlying mucosa was, however, ulcerated with surface gastric metaplasia, but no discrete foci of hetero-

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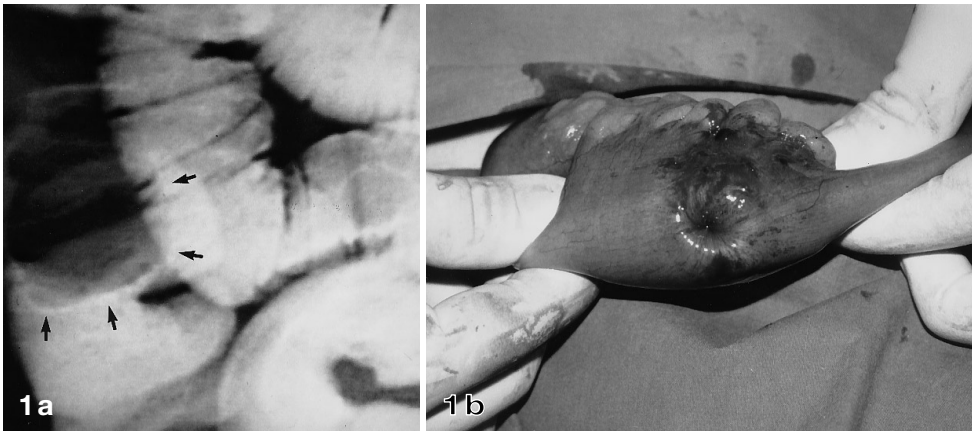


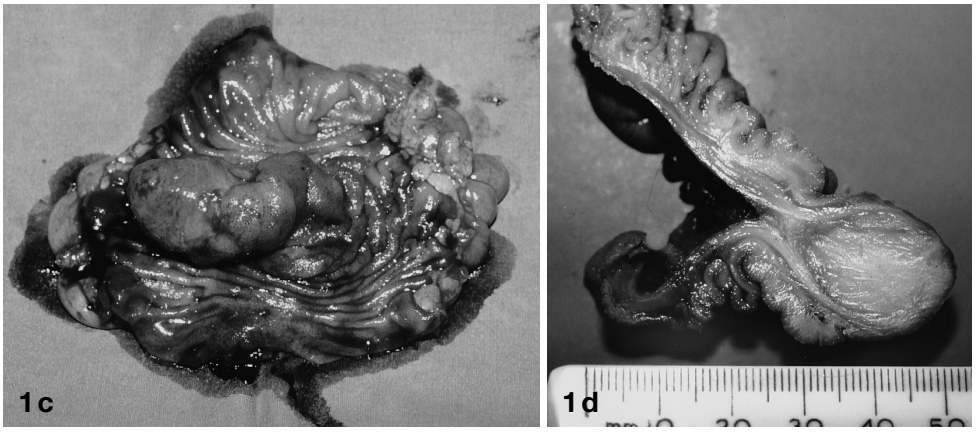
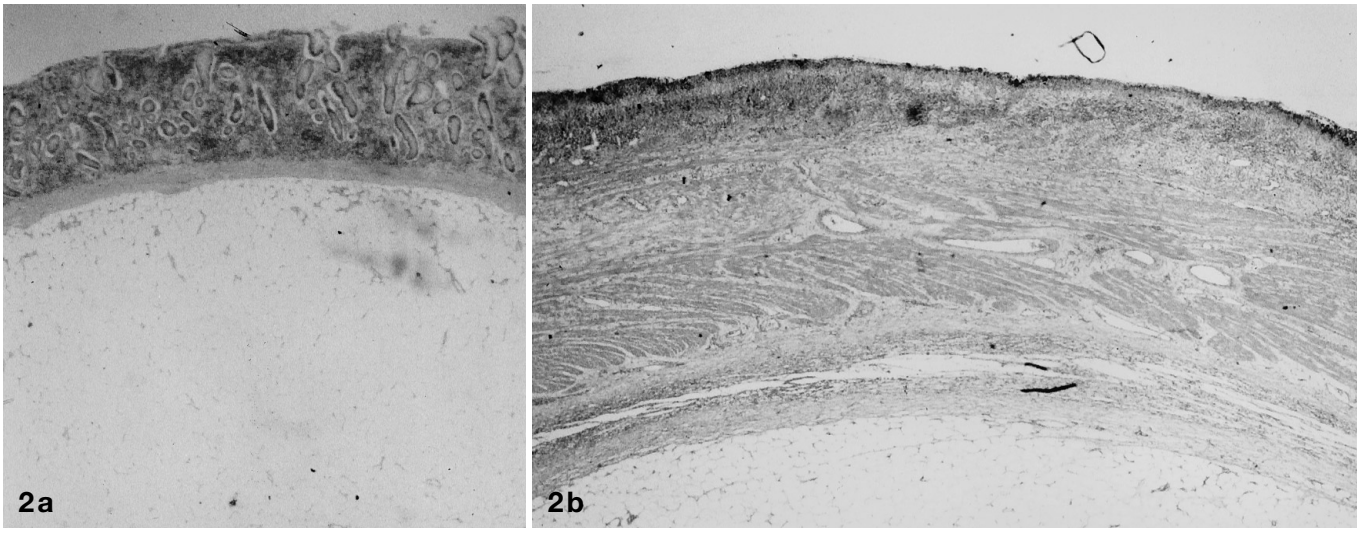


Fig. 1a–d. Case 1. **a** A small bowel enema (SBE) demonstrates a polyp in the distal ileum *(arrows)*. **b** Umbilicated lesion on antimesenteric border of ileal loop noted at surgery. **c** Resected small bowel opened to show a polypoid tumour with bulbous tip. **d** Resected specimen divided longitudinally to reveal a central mass of fat at the tip, simulating a lipoma

Fig. 2. Histological specimens, with the lumen uppermost, comparing **a** lipoma and **b** pseudolipoma of inverted Meckel's diverticulum (case 1). With the true lipoma, the adipose tissue is submucosal. The muscle layers and serosa (not shown) lie deep to the tumour. With the inverted Meckel's diverticulum, the adipose tissue lies deep to the full thickness of the bowel wall and is encapsulated by a second layer of serosa, with a gap between the two serosal layers



topic tissue. The final histological diagnosis was an isolated inverted Meckel's diverticulum.

Case 3

A 57-year-old man presented with a 1-month history of increasing shortness of breath on exertion and angina. There were no abdominal symptoms. Severe iron-deficiency anaemia (Hb 5.6 g/dl) was noted and faecal occult blood testing was positive. Upper gastrointestinal endoscopy, barium enema and colonoscopy were normal. An SBE revealed a polypoid filling defect in the ileum (Fig. 4 a). The lesion was identified with abdominal US (Fig. 4 b) and this demonstrated peristalsis of both the lesion itself and the small bowel loop in which it was contained. A CT examination identified central fat within the lesion (Fig. 4 c). Surgical findings were similar to case 1 and the lesion was resected (Fig. 4 d). Histology demonstrated subserosal fat. There was mucosal ulceration, but no discrete foci of heterotopic gastric mucosa. The final diagnosis was an isolated inverted Meckel's diverticulum.

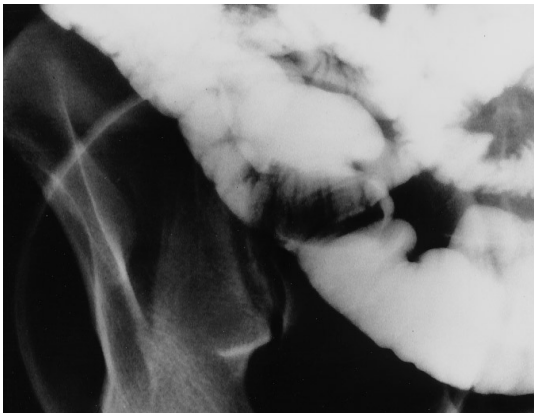


Fig. 3. Case 2. An SBE demonstrates a polyp in the distal ileum which was subsequently resected. Initial pathology reported a subserosal lipoma

Discussion

Meckel's diverticulum is the most prevalent congenital anomaly of the gastrointestinal tract and occurs in approximately 2% of the population [1–5]. The majority remain asymptomatic. The lifetime risk of Meckel's diverticulum causing disease is 4%, and this figure decreases with age [3]. Common complications include bowel obstruction, with or without intussusception, haemorrhage and diverticulitis [1–7]. Haemorrhage is most commonly due to peptic ulceration associated with heterotopic gastric mucosa, and presents most commonly in the paediatric age group [4, 6, 7].

Two large series reviewing 216 cases of intussuscepted Meckel's diverticulum have shown that virtually all cases are associated with invagination of the diverticulum, which is thought to act as the lead point [1, 2]. Peristalsis leading to intussusception after invagination is easily understood, but why invagination occurs in the first place is uncertain. It has been suggested that abnormal peristaltic movement due to ulceration or ectopic tissue at the base of the Meckel's diverticulum may cause it to invert [8]. This is not a unifying theory, as some reported cases demonstrated neither ulceration nor ectopic tissues [9, 10]. To our knowledge, it has never been suggested that invagination is congenital, although the evidence on which the acquired assumption is based is not clear. We are not aware of any reported case documenting a normal Meckel's diverticulum which has subsequently inverted.

An inverted Meckel's diverticulum without intussusception has rarely been documented. A recent review of 25 years of experience at the Armed Forces Institute of Pathology reported five such cases [11], but separate details of the individual cases were not recorded. We could only find another 12 case reports in the international literature [10, 12–21]. Three of these cases progressed to intussusception by the time of surgery [10, 17]. In two other cases abdominal pain was the predominant symptom [12, 15], but a coexisting intussuscepting jejunal polyp was present in one of these cases [15]. The remaining 7 cases (5 males and 2 females, mean age 48 years, range 26–78 years) all presented with anaemia and/or gastrointestinal bleeding [13, 14, 16, 18–21]. Abdominal pain was not a presenting symptom in these cases and in two the absence of pain was specifically recorded [13, 19]. Similarly, two of our three patients had no abdominal symptoms.

Most cases of isolated inverted Meckel's diverticulum have been associated with ulceration of the tip of the inverted diverticulum [13, 14, 16–19, 21], and this was present in all of our cases, which explains the clinical presentation of iron-deficiency anaemia and gastrointestinal bleeding. Ulceration may occur in the absence of heterotopic gastric mucosa [14, 18, 19], as occurred in two of the cases in this series. The explanation for this may be from chronic mechanical trauma or ischaemia from intermittent episodes of intussusception [19]. The latter would be symptomatic and both suggestions imply that ulceration occurs after invagination. Another explanation for ischaemic ulceration of a Meckel's diverticulum, whether inverted or not, lies in the nature of its blood supply. This is usually an end branch of the superior mesenteric artery – the persistent right vitelline artery – which does not anastomose with other ileal branches [7].

The radiological diagnosis of an inverted Meckel's diverticulum was made by barium examinations in 17 of 20 cases, including the three patients in this series. An SBE is considered to be the most dependable radiological method for demonstrating a Meckel's diverticulum [22], and this is also true when the diverticulum is inverted. Small bowel follow-through demonstrated five of eight cases [11, 13, 15–19, 21], whereas SBE was positive in all six cases [12, 18, 20] where the type of examination was recorded. Reflux of barium into the small intestine during a barium enema can also demonstrate an inverted Meckel's diverticulum [10, 12, 14, 17]. The inverted diverticulum is typically tubular, smooth and pliable, and may have a bulbous tip. A long tubular filling defect (case 3) is characteristic, but a shorter diverticulum (case 2) more closely resembles a polyp. Mucosal folds might be expected to be observed covering an inverted Meckel's diverticulum at enteroclysis, but in practice this has not been observed possibly because the mucosa is stretched or oedema effaces the folds.

Ultrasonic appearances of an intussuscepted Meckel's diverticulum have been described [11, 23], but those of an isolated inverted Meckel's diverticulum have not been previously reported. One important diagnostic feature was active peristalsis of the diverticulum during the ultrasound examination. The CT appearance has been reported previously [18] and the important observation is that of a central area of fat attenuation within the intraluminal mass. This represents the fat in the invaginated mesodiverticulum.

Radioisotope studies have a limited role in diagnosis. The literature records three cases in which pertechnetate scans were performed, and of these, two were normal; in both cases there was no ectopic gastric tissues present [10, 16]. In the other case [17], the abnormal pertechnetate scan was related to active bleeding, rather than uptake in ectopic tissue. Angiography was also ab-

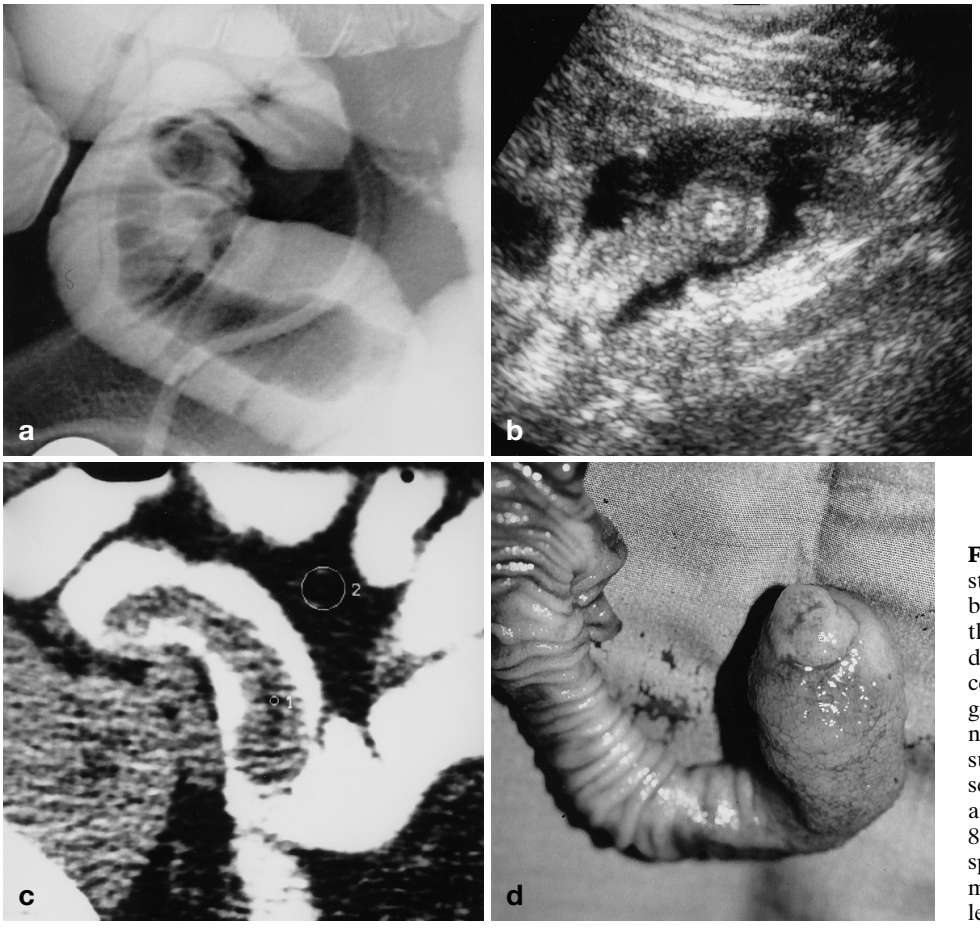


Fig. 4a–d. Case 3. **a** An SBE demonstrates a long tubular polyp with a bulbous tip. **b** A US examination identifies the intraluminal tumour. Real time US demonstrated active peristalsis. The central echogenic foci result from invaginated mesenteric fat. **c** A CT examination confirms the central adipose tissue with the same attenuation as mesenteric fat (regions of interest *circles 1* and *2* attenuation values were –87 and –89 HU, respectively). **d** The resected specimen. Notice that the small bowel mucosal folds are continuous with the lesion

normal in this case. In another case [19] a sulphur-colloid scan was negative, but a labelled red cell scan following this demonstrated active bleeding. Angiography in this case was negative.

The operative findings are of a tumour palpable within the distal ileum at which site a small umbilication is noted on the antimesenteric border of the gut (Fig. 1 b). A preoperative diagnosis of inverted Meckel's diverticulum can affect surgical management. In case 1 in this series, the surgeon was alerted to the potential diagnosis and opted to perform laparoscopy, confirmed the diagnosis by identifying the umbilication on the antimesenteric border of a loop of distal ileum and was then able to deliver the small bowel loop through a small appendicectomy-type incision. The small bowel could then be resected extra-abdominally with the dimple serving as a marker for the site of the tumour. Thus, the patient was saved from a standard laparotomy incision.

Histological examination of the inverted diverticulum reveals a variable amount of central adipose tissue [11, 14, 18, 20, 21]. This invaginated mesenteric fat is surrounded by a layer of mesothelial cells. In two of our cases this formed a discrete mass simulating a lipoma. However, the location of this adipose tissue is subserosal compared with the submucosal location of a true lipoma (Fig. 2). In one patient (case 2) the significance of this was not initially appreciated and in another (case 1) the significance was only realised after correlation with the operative findings.

Although an isolated inverted Meckel's diverticulum is apparently rare, it may be that some cases are overlooked without correlation of clinical, radiological and pathological findings.

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