Case report

Invasive amoebiasis: an unusual presentation

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SUMMARY  A 63 year old Asian woman who presented with three week’s abdominal pain was found to have a hard right iliac fossa mass and rectal ulceration. Profuse rectal bleeding necessitated a laparotomy. An inflammatory paracaelal mass with fistulae involving appendix, small bowel, and bladder was excised with exteriorisation of the bowel ends. Microscopy showed invasive amoebae. Re-anastomosis was successfully done after treatment with metronidazole and diloxanide. There are no previous reports of a paracaelal amoeboma with fistulae to either the appendix, or urinary bladder.

Amoebiasis is a protozoan infection caused by Entamoeba histolytica. It is often considered to be a tropical disease. The carrier rate in the United Kingdom, however, up to 1950 is reported as being 2–5% of the population, and does not appear to have been reappraised since.1-3 Carrier rates as high as 25% have been reported in some areas of the USA.4 The majority of carriers pass cysts in the stools but are otherwise asymptomatic.5 Approximately 5% of infected individuals develop clinical symptoms of amoebic dysentery† and of those requiring hospitalisation about 10% develop complications as a result of invasive amoebiasis.6

Complications which may arise include colonic perforation, liver abscess, pleural and pericardial effusions and enteric fistulae. The formation of an inflammatory mass or ‘amoeboma’ is another recognised but rare complication of invasive amoebiasis, most commonly affecting the caecum and presenting as a mass in the right iliac fossa. If treated promptly with chemotherapy complete resolution may be expected.7

We report a patient who developed a paracaelal amoeboma with fistulae to the appendix, terminal ileum and urinary bladder. A paracaelal amoeboma has not previously been reported with fistulae to either the appendix or urinary bladder.

Case history

A 63 year old Asian woman presented to the outpatient clinic with a three week history of anorexia, 6 kg weight loss, loose bowel motions, and right iliac fossa pain. Symptoms had started shortly before returning from a one month vacation in India. She had no significant past medical history and was not on any medication.

On abdominal examination a hard irregular mass was present in the right iliac fossa arising from the pelvis. Rectal examination and sigmoidoscopy revealed an irregular ulcer 4 cm above the dentate line which was biopsied.

On initial investigations the urea, electrolytes, glucose and amylase were normal. The alkaline phosphatase was 163 IU/l (normal range 40–130 IU/l), total protein 56 g/l (normal range 60–80 g/l), and albumin 19 g/l (normal range 35–55 g/l). The haemoglobin was 9.9 g/dl with normochromic and normocytic indices and the white cell count 14.5×10^9/l (neutrophils 81%, lymphocytes 11%, monocytes 5% – no eosinophilia).

Barium meal and follow through examination (Fig. 1) showed two areas of communication between the terminal ileum and a cavity in the right iliac fossa which was 8 cm in diameter on ultrasound examination, partly cystic, and partly solid. The right ureter was displaced laterally on an intravenous urogram. Stool microscopy showed no evidence of ova, cysts or parasites and rectal biopsy consisted of chronic inflammatory tissue with no diagnostic features.
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Fig. 1 Barium follow through examination showing communication between the terminal ileum and the right iliac fossa cavity.

She remained anorexic with a low grade pyrexia. Serum albumin concentration fell to 14 g/l and haemoglobin to 8.2 g/dl with a polychromic film.

The excised specimen (Fig. 2) consisted of terminal ileum, 19 cm long, caecum and lower descending colon, together measuring 8 cm in length, with attached appendix. The terminal ileum and the residual 2 cm stump of appendix lay within an opened abscess cavity, approximately 10 cm in maximum diameter. The lumen of the appendix stump and of the ileum freely communicated with the abscess cavity, there being an almost circumferential interruption in the continuity of the wall of the ileum in a 1 cm segment, 4 cm proximal to the ileocaecal valve.

Six shallow transverse ulcers, up to 2.5 cm across, covered by fibrinous exudate, were present in the caecal mucosa (Fig. 2). Histological examination of the edge of the ileal and appendiceal perforations and the lining of the abscess cavity showed granulation tissue, covered by fibrinous exudate. Haemophagous amoebae (Fig. 3) were present in the exudate in all areas, including the caecal ulcers, but no invasion of amoebae into otherwise intact bowel wall or involvement of blood vessels was found. It was concluded that the paracæcal amoebic abscess had originated from perforation of either the appendix or terminal ileum.

The patient was started on metronidazole and diloxanide and improvement in her condition was noted. Two weeks postoperatively she had another sudden and profuse fresh rectal bleed. After resuscitation and under general anaesthesia a colonic lavage was carried out through the mucous fistula allowing visualisation of the colon with the fibreoptic sigmoidsoscope. Several large ulcers were present in the lower rectum, one of which was actively bleeding. Under-running of the bleeding point per anally secured haemostasis.

Over the subsequent three weeks she began to eat and parenteral nutrition was discontinued. A cystogram showed satisfactory healing of the bladder and suprapubic drainage was successfully discontinued. She was allowed home.

Three months later colonoscopy confirmed healing of the amoebic ulceration. After ileo-colonic re-anastomosis she made an uneventful recovery.

Discussion

Despite the carrier rate for Entamoeba histolytica being at one time reported as high as 5% in the UK, only approximately 200 new cases of amoebiasis are now reported annually. This may reflect the difficulty of establishing a diagnosis with many cases either passing undetected or being misdiagnosed.

Stool analysis by standard methods is often unhelpful but serological testing for Entamoeba histolytica antigen by radioimmunoassay has been shown to be effective in all cases.

Complications occur when adequate supportive therapy and appropriate antibiotics are not initiated at an early stage. The main complications of colonic perforation and metastatic abscess formation are
associated with a high mortality whether managed conservatively or by surgery.\textsuperscript{11} A rarer complication of invasive amoebiasis is the formation of a chronic inflammatory mass or ‘amoeboma’. Amoebic granulomas may affect any part of the colon but are most common in the caecum, rectum, and transverse colon.\textsuperscript{12} In the present case an ‘amoeboma’ was present in the right iliac fossa. This was paracaecal in site and involved the appendix and terminal ileum. Amoebic ulceration was marked within the caecum. Two explanations may be postulated for the pathogenesis. First, this may have been a primary amoeboma of the appendix, a common site for amoebic ulceration but not previously reported as a site for granuloma formation. Alternatively necrosis of the appendix and terminal ileum may have occurred secondary to amoebic involvement of the regional blood vessels. It has been shown recently that perforation of the colon in amoebiasis is caused by vascular thrombosis secondary to direct spread of amoebae.\textsuperscript{13} No vascular invasion, however, was established on histological examination of regional vessels.

Complications which have been reported secondary to the development of an amoebic granuloma are ileocaecal intussusception, intestinal obstruction, and abdominal wall abscesses.\textsuperscript{12} There have been no previous reports of urinary fistulae secondary to amoebiasis although they are occasionally seen after acute purulent appendicitis.\textsuperscript{14} Involvement of the appendix rather than the caecum in granuloma formation may have predisposed to this complication.

Appropriate drug therapy is the key to the management of intestinal amoebiasis including granuloma formation. Emetine was the first effective treatment but has been superseded by metronidazole in acute invasive amoebiasis\textsuperscript{15} and diloxanide\textsuperscript{16} in chronic intestinal infections. In severe infections they may be used concurrently or sequentially. The role of surgery in amoebiasis is restricted to the emergency management of complications.
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Fig. 3  Section of the appendix wall showing haematophagous amoebae.

Before the advent of effective drug therapy post-operative complications such as exacerbations of colitis, haemorrhage, peritonitis and skin involvement were frequent resulting in surgical mortality rates of 25–40%. Bowel resections with anastomosis are associated with a high incidence of leakage and fistula formation. A satisfactory alternative, as utilised in the present case, is to resect the involved bowel and exteriorise the bowel ends. After cure of the amoebiasis and improvement in the general condition of the patient intestinal continuity may safely be restored.

References
