Colocutaneous fistula secondary to amoebiasis

Owain P. Jones*, John A. Murphy, Bushra N. Hamid, Dale Vimalachandran

Countess of Chester Hospital NHS Foundation Trust, Countess of Chester Health Park, Liverpool Road, Chester, Cheshire CH2 1UL, UK

Article info

Article history:
Received 9 December 2010
Received in revised form 10 December 2010
Accepted 13 December 2010
Available online 21 December 2010

Keywords:
Colocutaneous fistula
Appendicular abscess
Amoebic dysentery
Amoebiasis

Abstract

Here we present an interesting and extremely rare case of a 66 year old male who developed a colocutaneous fistula secondary to amoebiasis. The patient presented with an acute history of right lower abdominal pain, weight loss and a palpable mass. A CT scan demonstrated a fluid filled cavity in the right iliac fossa consistent with an appendiceal abscess which was drained under radiological guidance. However, following drainage his symptoms remained requiring open surgical drainage, and a controlled caecostomy was performed due to a small caecal perforation. Despite appropriate conservative therapy he failed to progress, and developed localised sepsis in the right iliac fossa with a colocutaneous fistula, requiring a formal right hemicolectomy. The histological examination confirmed the presence of abundant trophozoites of Entamoeba histolytica.

We highlight the fact that in the modern age of immigration and long distance travel, it will become increasingly likely that the so-called ‘tropical’ diseases will present throughout the world. This case also highlights the need to keep an open mind in cases that do not progress as expected, and to react accordingly to any unusual developments.

© 2010 Surgical Associates Ltd. Elsevier Ltd. All rights reserved.

1. Introduction

Entamoeba histolytica is particularly prevalent in tropical and subtropical regions, and is thought to affect 10% of the population, of which 90% will be asymptomatic. Colonic perforation is a widely known complication of the disease, though colonic fistulae are rare. We report a case of a colocutaneous fistula secondary to amoebiasis, which was initially managed as an appendiceal abscess.

2. Case presentation

We present an interesting case of a 66 year old gentleman with a history of hypertension. He was otherwise fit and well and had no previous abdominal surgery. He presented to the surgical department with a three week history of worsening right iliac fossa pain, more so over the previous week. There was no alteration in his bowel habit, but had lost a stone in weight over the preceding month.

On examination he appeared well, afebrile, with normal observations. The abdomen was soft with mild tenderness in the right iliac fossa where a mass was palpable. Per rectal examination was unremarkable. Initial haematological examination revealed Hb 11.6 g/dL, MCV 89 fL, WCC 9.9 × 10^9/L, urea, electrolytes and liver function tests were normal. C-reactive Protein was 199 mg/L and CEA 1.4 µg/L.

A CT scan revealed a focal fluid collection in the right iliac fossa measuring 5 cm × 5 cm × 7 cm (Fig. 1). The appendix was not easily identifiable, but a diagnosis of an appendiceal abscess was made. This was drained under radiological guidance due to his non-toxic status and he was discharged 48 h later with a drain in situ which was to be flushed daily.

On review five days later, his inflammatory markers were noted to be rising and a residual 6 cm × 4 cm abscess was identified on ultrasound examination. Intravenous Ertapenem was commenced and he was listed for surgical drainage. Under general anaesthetic a Lanz incision was made. A large abscess cavity was identified, as well as a small perforation noted in a friable caecum, which was presumed to be the site of the appendicular orifice. A Foley catheter was inserted and secured to act as a controlled faecal fistula (colocutaneous fistula). Post operatively the patient was commenced on total parenteral nutritional in order for spontaneous closure of the colocutaneous fistula.

Two weeks later, the catheter was draining minimal amounts, and both TPN and antibiotics were ceased. A fistulogram was performed with 50 ml of omnipaque contrast which showed the contrast to be confined to the lumen of the caecum and ascending colon (Fig. 2). The wound had become progressively necrotic, and this superficial necrosis was debrided in theatre. He was discharged home the following day with input from the tissue viability team.

Following a further two weeks of conservative therapy, the external opening of the colocutaneous fistula was noted to be...
enlarging with further loss of soft tissue causing local sepsis (Fig. 3), and a further CT scan revealed the colocutaneous fistula (Fig. 4) and marked thickening of the anterior abdominal wall consistent with infection.

In view of the apparent deterioration locally he underwent a laparotomy. Intraoperatively, a perforated caecum was noted to be fistulating into the anterior abdominal wall. It was presumed that a caecal malignancy was a strong possibility; however there was no evidence of metastases. A right hemicolectomy was performed followed by peritoneal lavage and a double barreled stoma was formed. Primary anastomosis was adjudged to be unsuitable given the patient's anaemia, low albumin and sepsis. Due to the extent of contamination of the right side of the abdominal wall, this stoma was sited in the left iliac fossa. Following colonic resection he made an uneventful recovery.

Histopathological examination of the caecum revealed active inflammation and flask shaped ulceration of the mucosa associated with full thickness necrosis resulting in caecal perforation. The inflammatory exudate and the ulcerating tissue contained abundant trophozoites of *E. histolytica* (Fig. 5). The bowel mucosa away from the ulcerated area was unremarkable. On further questioning the patient recalled that he had relatively recently (within the last twelve months) visited Cape Verde and had developed a short 'Gastroenteritis' like illness there which had settled with no specific intervention. He was commenced on a course of metronidazole, and an ultrasound of his liver excluded the presence of abscesses. Five months later his wound had fully epithelialised and the stoma has been successfully reversed.
3. Discussion

It is estimated that 10% of the world’s population is infected with *E. histolytica*, with 90% of those affected being asymptomatic. The World Health Organisation suggests that this pathogen is responsible for 70,000 deaths annually, and is second only to malaria as the leading cause of death from a parasitic infection worldwide. *E. histolytica* is particularly prevalent in tropical and subtropical regions, and the disease is transmitted via the ingestion of cysts, usually via faecally contaminated food or water.

A variety of diagnostic tests can be utilised in the identification of *E. histolytica*, and their success rates are dependent on whether the patient is asymptomatic, or suffering from intestinal or extra-intestinal disease. In those with amoebic colitis, stool microscopy in less than 60% sensitive and 10–50% specific. In contrast stool antigen detection (ELISA) is over 95% sensitive and specific, with other less accurate methods available including serum antigen detection, serum antibody detection, and stool PCR.

Invasive infection with the protozoan *E. histolytica* commonly causes diarrhoea with blood and mucus, abdominal pains and fever. This myriad of symptoms is known as amoebic colitis and its symptoms can alternate with episodes of constipation, or even remission. This anaerobic parasite lives in the colon as a harmless organism, and can lie dormant for many years following exposure. What triggers this conversion into an invasive, clinically apparent disease in currently unknown.

Intestinal sequelae of the disease include toxic megacolon, which develops in 0.5% of those suffering from colitis. Colonic ulceration can also occur, which if left to progress can lead to visceral perforation as demonstrated in our patient. This complication has an estimated prevalence of up to 6% in those suffering from amoebiasis. In one Mexican study of 122 amoeba-related colonic perforations, over 90% were localised to the right colon, with multiple perforations evident in 74% of cases. 1 in 3 patients undergoing a right hemicolecotony in this group died, with a general mortality rate of 40% in the entire patient cohort. A Japanese series reports a mortality rate of 92% in those undergoing resection for colonic perforation, though it must be emphasized that both these series span 1970s–1990s and significant progress has been made since in diagnostic techniques, surgical techniques and critical care.

Various surgical approaches have been described in the literature in the management of perforated amoebic colitis depending on the degree of peritoneal contamination. Adams et al. suggest that perforation occurs either acutely or as a microperforation, the latter of which can be treated conservatively. However, surgery is certainly indicated where there is evidence of gross peritoneal contamination. Procedures described include resection, diverting stomata, and primary closure of the perforation. Primary anastomosis is contraindicated in the presence of amoebiasis, even in the absence of any contamination. The authors could only identify one colocutaneous fistula as a result of amoebic colitis which was over a quarter of a century ago. Gupta describes sigmoid amoebiasis with pericolic abscess formation which ruptured externally causing a spontaneous fistula.

Several other intestinal complications have been described as a result of *E. histolytica*. Fistulae, including rectovaginal, enterohepatic, and cholecystocolonic, and colonic strictures, and rarely, a granulomatous pseudotumour known as an ameboma can develop, potentially causing large bowel obstruction or intussusception.

Extra-intestinal disease commonly involves the liver due to the portal venous drainage of the colon. Liver abscesses are common, which can rupture intraperitoneally leading to peritonitis. Hepatic abscesses can also fistulate superiorly, either into the pleural or pericardial cavities, depending on which lobe of the liver is mainly affected. Other manifestations of the disease include cerebral abscesses, genitourinary involvement, and cutaneous disease.

4. Conclusion

What was originally thought to be a routine appendiceal abscess certainly did not follow the expected path to recovery. A factor
which made this case a diagnostic challenge was the absence of diarrhoea. This symptom alone could potentially have resulted in an earlier diagnosis, either through increased clinical suspicion, or by the identification of trophozoites from a stool sample. It is difficult to speculate whether earlier diagnosis could have led to successful conservative management, but we acknowledge that is a possibility.

In the modern age of immigration and long distance travel, it will become increasingly likely that these so-called ‘tropical’ diseases will be presenting in the unlikeliest of places. This case highlights the need to keep an open mind in cases that do not progress as expected, and to react accordingly to any unusual developments.

Conflict of interest

None

Funding

None

Ethical approval

None

References

1. Lacasse A, Cleveland KO. Amebiasis. eMedicine 2009. Updated: February 9.
2. WHO. Life in the 21st Century: a vision for all. The World Health Report 1998. Geneva, Switzerland: World Health Organization; 1998.
3. Stanley Jr SL. Amoebiasis. Lancet 2003;361(9362):1025–34.
4. Nuran D, Gonul A, Mehmet S, Cahit B, Arzu K, Gurrol E. Detection of entamoeba histolytica/entamoeba dispar in stool specimens by using enzyme-linked immunosorbent assay. Mem Inst Oswaldo Cruz 2004;99(7):769–72.
5. Tanyuskel M, Petri Jr WA. Laboratory diagnosis of amebiasis. Clin Microbiol Rev 2003;16(4):713–28.
6. Dans LF, Martínez EG. Amebic dysentery. Clin Evid (Online) 2007;2007:0918.
7. Pritt BS, Clark CG. Amebiasis. Mayo Clin Proc 2008;83(10):1154–9.
8. Mortimer L, Chadee K. The immunopathogenesis of Entamoeba histolytica. Exp Parasitol 2010;126(3):366–80.
9. ChenWJ, Chen KM, Lin M. Colon perforation in amoebiasis. Arch Surg 1971;103:676.
10. Achié-Gutiérrez C, Rodia-Rosas H, Guizar-Bermúdez C, ALCÁNTARA A, Montalvo-JáVÉ EE. Evolution of surgical treatment of amebiasis-associated colon perforation. J Gastrointest Surg 2010;14(1):82–7.
11. Nishiwaki N, Honda K, Kishikawa H, Tanaka H, Taniwaki S, Naruse H, et al. Two cases of amebic colitis associated with colon perforation (in Japanese with English abstract). Nihon.
12. Adams EB, MacLeod IN. Invasive amebiasis. I. Amebic dysentery and its complications. Medicine 1977;56:315–23.
13. Ishida H, Inokuma1 S, Murata1 N, Hashimoto1 D, Satoh K, Ohta S. Fulminant amebic colitis with perforation successfully treated by staged surgery: a case report. J Gastroenterol 2003;38:92–6.
14. Gupta S. Spontaneous colcutaneous fistula in amoebic colitis. Trop Geogr Med 1983;35(3):305–7.
15. Loprinzi C, Heaton JW, Kelly PC. Enterohepatic fistula associated with amebic liver abscess. SMJ 1983;76(3):384–5.
16. Menda RK, Chulani HL. Cholecystocolonic fistula following ameboma of the ascending colon: report of a case. Dis Col Rectum 1971;14(5):386–8.
17. Cain GD, Wolma Jr FJ. Patterson M. Extensive stenosis of the colon and fistula formation following amoebic dysentery. Gastroenterology 1971;61(8):898–900.
18. Medical microbiology. 4th ed. University of Texas Medical Branch; 1996.
19. van Hal SJ, Stark DJ, Fotedar R, Marriott D, Ellis JT, Harkness JL. Amebiasis: current status in Australia. MJA 2007;186(8):412–6.