

First case of villous adenoma of the appendix leading to acute appendicitis presenting as strangulated femoral hernia: changes in management owing to concurrent adenoma

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Abstract A 78-year-old lady presented with signs and symptoms of a strangulated femoral hernia. Peri-operatively she was found to have appendicitis within the hernia sac. Appendicectomy and non-mesh hernia repair were performed. Histology revealed acute inflammation and a villous adenoma of the appendix. Villous adenomas of the vermiform appendix are extremely rare tumours of the gastrointestinal tract. This is the first case combining two very rare pathologies—acute appendicitis presenting as strangulated femoral hernia and villous adenoma of the appendix. Early diagnosis and surgery are required to avoid high morbidity of perforated appendicitis within a femoral hernia. First, we discuss the diagnosis and surgical treatment of acute appendicitis within a femoral hernia. Second, the presence of an adenoma changes the aetiology of appendicitis. More importantly, changes in surgical management of acute appendicitis presenting as a strangulated femoral hernia owing to a co-existing adenoma are discussed.

Keywords Appendicitis · Femoral hernia · Appendix tumour · Benign · Adenoma · Treatment

Introduction

Femoral herniae are among the least common groin herniae, accounting for fewer than 10% of groin herniae and

3% of all abdominal wall herniae [1, 2]. The vermiform appendix is rarely found within the femoral hernia but even more rare is the development of acute appendicitis within the femoral hernia which then presents as a strangulated femoral hernia [2]. Early recognition expedites diagnosis and surgery in this serious condition. Villous adenomas of the vermiform appendix are extremely rare tumours of the lower gastrointestinal tract. Only 43 cases have been described [3]. We report the first case combining these two extremely rare pathologies—acute appendicitis presenting as strangulated femoral hernia and villous adenoma leading to appendicitis. We discuss the diagnosis and surgical treatment of acute appendicitis presenting as strangulated femoral hernia. We also discuss how the presence of a villous adenoma alters the pathogenesis and surgical management of patients with appendicitis presenting as strangulated femoral herniae.

Case report

A 78-year-old Caucasian lady presented with a two-week history of intermittent right groin swelling and lower abdominal pain after a bout of coughing. There were no symptoms of obstruction. Examination revealed a tender, irreducible mass in the right groin with no evidence of peritonitis. Blood tests showed haemoglobin of 12.6 g dL⁻¹ (normal 11.5–16.5 g dL⁻¹) and mild leukocytosis 12.8 × 10⁹ L⁻¹ (normal 4–11 × 10⁹ L⁻¹) with neutrophilia 10.2 (2–7.5 × 10⁹ L⁻¹). A diagnosis of strangulated femoral hernia was made and an incision made over the site of the suspected hernia rather than a midline incision, because there was no evidence of bowel necrosis or peritonism. Peri-operatively the femoral hernia was found to contain an inflamed appendix which required extension of the incision

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to obtain peritoneal entry and control of the appendix base. Appendectomy was performed by transfixing the base of the appendix and burying the appendiceal stump. Closure was performed using monofilament polydioxanone (PDS; Ethicon, Johnson&Johnson Healthcare Systems, USA) to the peritoneum and non-absorbable monofilament polypropylene (Prolene; Ethicon, Johnson&Johnson Healthcare Systems) to oppose the iliopubic tract to Cooper's ligament. A mesh repair was felt inappropriate, because of the risk of infection in the presence of inflammation and appendix excision. She was discharged four days later. Histology confirmed acute appendicitis but further revealed a villous adenoma of the appendix which was confined to the mucosa and not extending to resection margins (Fig. 1). As a result she had a barium enema which showed normal appearances of the remaining large bowel. She remains well 20 months later.

Discussion

Femoral herniae are rare and account for 3% of all abdominal wall herniae [1, 2]. Despite this, it has the highest morbidity and mortality in emergency hernia surgery [4]. This is partly because of the high cumulative strangulation rates of 22% at three months and 45% at twenty-one months [5]. Anatomically, the narrow femoral neck leads to strangulation of the hernia sac contents which often include omentum or small bowel. The appendix is found incidentally within a femoral hernia on rare (<1%) occasions, most commonly during elective repair [6]. Even rarer is appendicitis which develops within the hernia and then subsequently presents as a strangulated femoral hernia. Since its first description by De Garengot in 1731, only 72 cases (including this case) have been reported [7, 8].



Fig. 1 Appendix showing acute inflammation of the appendix (A) with villous adenoma of appendix (B) and intact muscularis mucosa (C)

Diagnosis

Acute appendicitis within a femoral hernia occurs most commonly in women and with increasing age. Both these factors are consistent with the population most likely to develop femoral herniae. A study of appendiceal perforations within femoral herniae showed a female preponderance (77% women compared with 23% men) with a mean age of 69 years [2]. Symptoms can be acute or chronic ranging from 2 days to 15 years [2]. It most commonly presents as a strangulated femoral hernia but can present as necrotising fasciitis, a groin mass, or small bowel obstruction [7, 9]. A large proportion of patients do not display cardinal features of appendicitis, for example pain, pyrexia, nausea, vomiting, anorexia, peritonism and high WBC counts, however [10, 11]. This is especially true in the elderly, in which this condition most frequently presents. In a series of patients >60 years with appendicitis fewer than 20% had classical signs of anorexia, pyrexia, right quadrant pain, or elevated white cell count [12]. Similarly, the patient in our case was afebrile with borderline leukocytosis and neutrophilia. There were no systemic features of appendicitis or peritonism on examination. The reason for symptom masking is probably the tight hernial neck which contains the perforated appendix within the sac thus preventing more widespread inflammation and peritonism [2]. The combination of under-diagnosis of femoral hernia, absence of signs of appendicitis in the elderly, and symptom masking because of the tight hernia neck leads to delay in diagnosis and surgery which results in greater occurrence of perforation and mortality, especially in the elderly [7].

Clinical features, for example an atypical history, subcutaneous air and induration over the hernia, crampy rather than constant pain, localised tenderness over the hernia, and the presence of radiological features of small bowel obstruction with dilated loops or air-fluid levels history, may aid diagnosis of underlying perforated appendicitis in patients with femoral hernia [7, 10]. Contrast-enhanced computed tomography (CT) is highly sensitive and specific and has been used to diagnose femoral appendicitis preoperatively but the rarity of this condition makes this infeasible for routine use, as it was in our case [13].

Overall villous adenomas account for only 12% of all colorectal adenomas. Villous adenoma of the vermiform appendix is an extremely rare tumour. Only 43 cases have been reported. Jafari et al. [14] further suggest that only 12 of the 43 cases have adequate clinical and pathological data to support this diagnosis. There is no sex predominance and the average age of presentation is 45 years (range 17–80 years). It most commonly presents with acute appendicitis or appendicular symptoms, which occur in 54.3% of cases [3]. The others are found incidentally, although some may present as intussusception [15].

Pathogenesis of acute appendicitis: intraluminal obstruction versus extraluminal constriction

The proposed aetiology of intra-abdominal appendicitis is different from that of acute appendicitis within a femoral hernia. The former is thought to be a result of intraluminal obstruction (e.g. lymph node hypertrophy or faecolith) leading to mucosal and transmural inflammation, necrosis, and perforation [16]. Conversely, appendicitis within a femoral hernia is more likely to be because of extraluminal compression caused by the narrow hernial neck resulting in inflammation and perforation [7, 9]. Our case is unique, however, because of the simultaneous presence of an appendix adenoma, which most commonly presents as intra-abdominal appendicitis [3]. Hence, in contrast with other cases of appendicitis within a femoral hernia, the adenoma in our case supports the intra-abdominal/intraluminal sequence of events i.e. intermittent intraluminal inflammation caused by the adenoma leading to acute appendicitis and inflammation of the hernial sac.

Peri-operative management

Appendicitis is diagnosed peri-operatively because patients often lack clinical signs and femoral hernia strangulation is an immediate indication for surgery. Appendectomy and femoral hernia repair are performed. The choice of tissue or prosthetic mesh repair depends on the surgeon. Prosthetic mesh can be used when there is no evidence of perforation. Mesh infection occurs even with a non-inflamed appendix, however [8]. Appendix perforation is a contraindication to mesh use because of risk of infection. Our surgeon performed tissue repair by suturing the iliopubic tract to Cooper's ligament. Antibiotic use should be considered in all cases, especially if prosthetic mesh is used or perforation is present. A meta-analysis of 2,532 patients found a fivefold increase in wound infection in perforated appendicitis within a hernia sac compared with intra-abdominal appendix perforation [17]. This is probably because of the delay in diagnosis, entry of multiple tissue planes, and poor patient status. Finally, if perforation is present, use of a groin incision only may prevent intra-abdominal dissemination of infection [18].

Although villous adenoma of the appendix is almost always diagnosed post-operatively, peri-operative features suggestive of a tumour include an irregular mass lesion, hernia irreducibility, inflammatory adhesions, or direct sac invasion. Caecostomy is advised for direct examination if caecal mass/extension is suspected. The choice of performing an appendectomy and awaiting histology (with or without interval right hemicolectomy) or immediate right hemicolectomy depends on the strength of suspicion of malignancy, the size of the tumour, and the ability to obtain

definite macroscopic clearance margins with simple appendectomy or invasion of the base of the appendix/caecum which would necessitate bowel resection.

Post-operative follow-up

The presence of an adenoma on post-operative histology changes the management of patients with acute appendicitis within a femoral hernia. These are regarded as pre-malignant and up to 29% may already contain invasive cancer [19]. The rate of malignant transformation is variable ranging from 6.2 to 75% [20]. This wide range reflects the limited information available about this tumour, because of its rarity. Clinically, if the adenoma does not extend beyond the submucosal layer or resection margins, simple appendectomy with femoral hernia repair is sufficient. If, however, the tumour extends submucosally or into the resection margins, interval right hemicolectomy should be performed.

Conclusion

We present the first case combining two extremely rare clinical entities. First, acute appendicitis presenting as strangulated femoral hernia. Strong suspicion is required, because signs of appendicitis are usually absent and delayed surgery is associated with increased morbidity and mortality. Several clinical features and CT imaging may aid diagnosis. Appendectomy and hernia repair should be performed but the risk of prosthetic mesh infection should be considered and routine use of antibiotics must be considered. Second, the coexisting presence of a tumour changes the possible pathogenesis of this condition. More importantly, all patients require lower gastrointestinal evaluation and some may require further surgery.

References

1. Devlin H (1982) Hernia. In: Russell RCG (ed) Recent advances in surgery. Churchill Livingstone, London, pp 54
2. Nguyen ET, Komenaka IK (2004) Strangulated femoral hernia containing a perforated appendix. *Can J Surg* 47:68–69
3. Lall KS, Mavrelis WP (1982) Villous adenoma of the appendix: report of a case and review of the literature. *Dis Colon Rectum* 25:716–719
4. Brittenden J, Heys SD, Eremin O (1991) Femoral hernia: mortality and morbidity following elective and emergency surgery. *J R Coll Surg Edinb* 36:86–88
5. Gallegos NC, Dawson J, Jarvis M, Hobsley M (1991) Risk of strangulation in groin hernias. *Br J Surg* 78:1171–1173
6. Wyatt JP, Varma JS (1992) Femoral hernia appendix causing small intestinal obstruction. *Postgrad Med J* 68:223–224
7. Isaacs LE, Felsenstein CH (2002) Acute appendicitis in a femoral hernia: an unusual presentation of a groin mass. *J Emerg Med* 23:15–18

8. Akopian G, Alexander M (2005) De Garengeot hernia: appendicitis within a femoral hernia. *Am Surg* 71:526–527
9. Guirguis EM, Taylor GA, Chadwick CD (1989) Femoral appendicitis: an unusual case. *Can J Surg* 32:380–381
10. Griffin JM (1968) Incarcerated inflamed appendix in a femoral hernia sac. *Am J Surg* 115:364–366
11. Serrano A, Ackerman NB (1979) Perforated appendicitis in an incarcerated inguinal hernia. *Arch Surg* 114:968
12. Horattas MC, Guyton DP, Wu D (1990) A reappraisal of appendicitis in the elderly. *Am J Surg* 160:291–293
13. Zissin R, Brautbar O, Shapiro-Feinberg M (2000) CT diagnosis of acute appendicitis in a femoral hernia. *Br J Radiol* 73:1013–1014
14. Jafari N, Christ M, Menaker GJ, Sheridan JT (1979) Villous adenoma of appendix—report of a case and review of the literature. *Am J Proctol Gastroenterol Colon Rectal Surg* 30:30–32
15. Tonsekar KS, Cacadac R, Ashare R, Libcke JH (1994) Villous adenoma of the vermiform appendix with cecal intussusception: a case report and review of literature. *Am Surg* 60:982–984
16. Temple CL, Huchcroft SA, Temple WJ (1995) The natural history of appendicitis in adults. A prospective study. *Ann Surg* 221:278–281
17. Rucinski J, Fabian T, Panagopoulos G, Schein M, Wise L (2000) Gangrenous and perforated appendicitis: a meta-analytic study of 2532 patients indicates that the incision should be closed primarily. *Surgery* 127:136–141
18. Barbaros U, Asoglu O, Seven R, Kalayci M (2004) Appendicitis in incarcerated femoral hernia. *Hernia* 8:281–282
19. Welch JP, Welch CE (1976) Villous adenomas of the colorectum. *Am J Surg* 131:185–191
20. Goligher JC (1984) Surgery of the anus, colon and rectum. In: Goligher JC (ed) *Surgery of the anus, rectum and colon*. Balliere Tindall, London, pp 437