Case Report

A rare presentation of xanthogranulomatous appendicitis and caecal angiolipoma in the same patient

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Abstract
In this communication, we report on a 49-year-old male patient who presented with acute appendicitis and appendicular mass which was treated conservatively. However, his condition deteriorated and appendicular abscess was suspected. He underwent surgical exploration. A large hard and gelatinous appendicular mass was found, which proved to be xanthogranulomatous appendicitis. However, the histopathological appearance of the xanthogranulomatous appendicitis was confused with pseudomyxoma peritonei and signet ring carcinoma of the appendix. Based on that, he underwent right hemicolecotomy. Incidental finding was a caecal angiolipoma. A meticulous literature search showed no reported combination of these two conditions in the same patient.

Keywords: angiolipoma, caecum

Case Report
The patient is a 49-year-old male from Central Sudan who presented to the Accident and Emergency Department with right lower quadrant abdominal pain. His condition started three days prior to presentation with vague lower abdominal pain. It was more pronounced on the right iliac fossa and was aggravated by coughing and movement. It increased gradually in severity and was associated with nausea, vomiting and anorexia. He is known to have mitral valve disease and was on aspirin 75mg and bisoprolol 0.5mg O.D. He had a past history of left ureteric colic caused by left ureteric stone. Systemic enquiry, drug, family or social history was not contributory to his presentation.

On examination, the patient was ill and in pain. He was not pale, jaundiced or cyanosed. His temperature was 37ºC. Systemic examination was within normal apart from evidence of mitral valve disease. Local abdominal examination revealed a scaphoid abdomen. There was guarding and rebound tenderness on deep palpation. A
A palpable mass was felt in the right iliac fossa. The psoas sign was positive. His renal, hepatic function tests as well as the haematological profiles were within normal limits. Abdominal ultrasound examination showed a mass in the right iliac fossa suggestive of an appendicular mass. The patient was treated conservatively in the form of intravenous fluids, ceftriaxone and metronidazole for 10 days. His general condition suddenly deteriorated and he developed evidence of peritonitis. An abdominal ultrasound and CT scan examinations showed evidence of early abscess formation in the right iliac fossa lateral to the psoas muscle and an appendicular mass. He underwent exploration. A large hard and gelatinous appendicular mass was found. It was tightly adherent to the caecum and terminal ileum. Appendicectomy was performed with uneventful post-operative recovery. The histopathological examination of the appendix showed a chronic inflammatory reaction composed of lymphocytes, plasma cells, and few neutrophils. The inflammation involved all the layers locally and was associated with mucin dissecting the muscles. There were several granulomas composed of foreign body giant cells, in line with the diagnosis of xanthogranulomatous appendicitis. However, it was incorrectly diagnosed by other pathologist as pseudomyxoma peritonei and signet ring carcinoma of the appendix. (Figs 1, 2, 3).

Colonoscopy to assess the intraluminal extension of the pathology was done and it was normal apart from evidence of extraluminal caecal compression. The patient had an abdominal CT scan which showed the presence of a caecal mass and left hydronephrosis due to a ureteric stone (Fig 4).
The patient underwent right hemicolectomy with uneventful post-operative recovery. The histopathological examination of the surgical specimen revealed evidence of angiolipoma of the caecum (Fig 5).  

Fig 5: Part of the angiolipoma of the cecum in the submucosa. (H&E x 40)  

Discussion  
Xanthogranulomatous inflammation is a rare pathological condition. It had been reported in many organs, most frequently in the kidney (1) and gallbladder. (2) There have been few reports of its occurrence in the appendix. (3) Appendicular histopathological examination in patients who undergone delayed appendicectomy have shown a strong association with granulomatous and xanthogranulomatous appendicitis. It was postulated that, the ruptured acute appendicitis which is often treated with antibiotic therapy and interval appendectomy may develop granulomatous appendicitis. The reported patient, did not respond to antibiotics and had ruptured his appendix; and that was probably the cause for the of the xanthogranulomatous appendicitis. (4) It was reported that, delayed or interval appendicectomy specimens often have a characteristic inflammatory pattern that includes granulomas, xanthogranulomatous inflammation, mural fibrosis and thickening, and transmural chronic inflammation. These changes may be misinterpreted for Crohn’s disease. In the reported patient there was neither clinical nor histopathological evidence of Crohn’s disease in the small or large bowel. (5)  
Angiolipoma is benign neoplasm with characteristic vascular pattern that is frequently seen in the subcutaneous tissue but rarely reported in the gastrointestinal tract. (6) In the gastrointestinal tract they are usually asymptomatic but abdominal pain, bleeding and obstruction may be the presenting symptoms in some patients. (7) As far as we are aware only two cases of angiolipoma localised on the ileocecal valve were previously reported. (5,8) In the reported patient, the angiolipoma was a histopathological surprise as the CT scan findings were not specific. Although it is a rare entity it should be considered in the differential diagnosis of a caecal mass. In appendicular masses careful intra-operative examination of the terminal ileum and caecum is important to avoid missing such double pathology. It is interesting to note that, the histopathological appearance of the Xanthogranulomatous appendicitis in this patient was confusing and it was reported by other pathologist as pseudomyxoma peritonei and signet ring carcinoma of the appendix.
References