Case Report

Xanthogranulomatous appendicitis mimicking appendicular lump: An uncommon entity

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ABSTRACT

Xanthogranulomatous inflammation is rare, and though it has been described in many organs, most commonly it occurs in the kidney and gall bladder. Xanthogranulomatous appendicitis is a very rare phenomenon. Clinical findings and imaging modalities are not sufficient for definite preoperative diagnosis. Clinical and radiological resemblances of these lesions to neoplastic processes often warrant excision. A 40-year-old female presented with pain in the right lower quadrant of the abdomen, loss of weight, nausea, and abdominal distension since 15 days. Ultrasonography and computed tomography of the abdomen and pelvis suggested neoplastic lesion. Surgical resection of the ileum, ileocecal junction with appendix, and part of transverse colon were done. Diagnosis of xanthogranulomatous appendicitis was made on histopathology examination.

Key words: Appendix, histiocytes, inflammation, xanthoma

INTRODUCTION

Xanthogranulomatous inflammation is a rare form of chronic inflammation and is a well-defined entity. Though it has been described in many organs, most commonly it occurs in the kidney and gall bladder. Xanthogranulomatous inflammation of appendix is a very rare phenomenon with few cases reported in literature.[1-4] It may mimic a locally advanced cancer. Imaging modalities are nonspecific. Definite diagnosis requires histopathological examination. We report a case of 40-year-old female presenting with appendicular lump.

CASE REPORT

A 40-year-old female presented with pain in a right lower quadrant of the abdomen, loss of weight, nausea, and abdominal distention since 15 days. There was no history of fever, constipation, and vomiting. She was P3 L3 and gave history of lower segment cesarean sections in all deliveries. There was no other significant medical history. On clinical examination, the abdomen was distended (Girth - 87.5 cm), soft, and tender all over. No guarding rigidity and rebound tenderness. Peristalsis was not seen. There was no hepatomegaly or splenomegaly or ascites. Scar of lower cesarean section was seen.

Ultrasonography (USG) of abdomen and pelvis revealed loculated anechoic collection in the right inguinal fossa surrounding bowel loops with thickening of ileocecal junction. Infective or neoplastic processes of bowel were suspected. A differential diagnosis of the right ovarian simple cyst was also suspected. Computed tomography (CT) of abdomen and pelvis showed hypo dense multiloculated collection in right iliac fossa along with cecum with thickened terminal ileum, mostly appendicular lump [Figure 1a]. Colonoscopy showed irregular thickening and ulceration at the base of appendix. Neoplastic pathology was suspected. Multiple biopsies from ileocecal junction did not reveal either inflammatory or neoplastic pathology.

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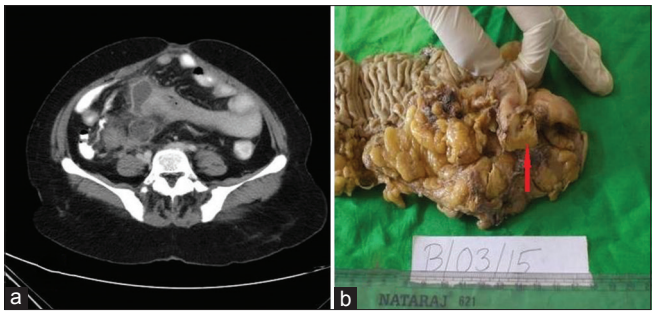

Figure 1: (a) Computed tomography abdomen and pelvis. (b) Specimen of ileocecal junction and thick walled appendix with yellowish deposits (arrow)

A surgical specimen of ileum measuring six cm, ileocecal junction with already cut appendix, and part of transverse colon was received for histopathological examination. Cut section of appendix showed thickened wall with yellowish white deposits [Figure 1b arrow]. Ileum and colon did not show any pathology grossly.

Histopathological sections from appendix showed focal ulcers in the mucosa. It was infiltrated by polymorphs and lymphocytic inflammatory infiltrate. The submucosa and serosa showed clusters of lipid-laden histiocytes (xanthoma cells) mingled with polymorphs, lymphocytes, plasma cells, and multinucleated giant cells [Figure 2a-c]. Similar cells were seen in serosa. Epithelioid granulomas or Michaelis–Gutman bodies were not seen. Sections from ileum and colon did not show significant pathological findings. A diagnosis of xanthogranulomatous appendicitis was made.

DISCUSSION

Xanthogranulomatous inflammation is a rare but a well-defined entity. It was first described in the kidney by Osterlind in 1944. Involvement of appendix is a rare phenomenon with few cases reported in the literature.[1-8] Birch *et al*. published first report xanthogranulomatous appendicitis in 1993.[1] The proposed causes are obstruction, hemorrhage, inflammation, local hypoxia, and perforation. In our case, inflammation can be the possible cause. The proposed pathogenic mechanisms include defective lipid transport, immunological disturbance, and infection by low virulence organisms.[5] It is generally believed that the localized proliferation of lipid-laden foamy histiocytes represents chronic suppurative inflammation which is secondary to interaction between host and microorganisms.[4,5]

Clinical presentation in xanthogranulomatous appendicitis is quite variable. Patients usually present with right lower abdominal quadrant pain, fever, nausea, and vomiting. Birch *et al*. suggested an association of the xanthogranulomatous response with long-standing inflammation of the appendix and formation of the appendiceal mass.[1]

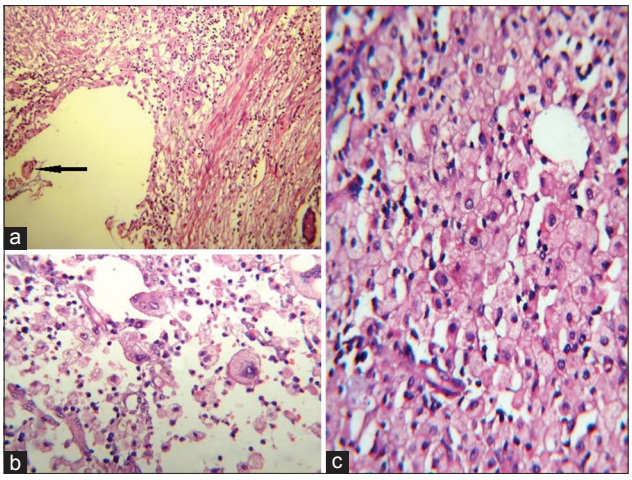

Figure 2: Sections from appendix show (a) submucosal collection of foamy histiocytes and giant cells (arrow, H and E, ×100). (b) Foamy histiocytes and multinucleate giant cells admixed with acute and chronic inflammatory cells (H and E, ×400). (c) Solid sheets of foamy histiocytes (H and E, ×400)

In contrast, Munichor *et al*. and Omar *et al*. reported cases of xanthogranulomatous appendicitis with typical signs and symptoms of acute appendicitis.[3,9] Chuang *et al*. reported a case of xanthogranulomatous appendicitis which mimicked like a locally invasive cancer.[8]

Imaging modalities are nonspecific.[6] In our case, USG suggested infective or neoplastic pathology. CT scan suggested the diagnosis of appendiceal tumor.

Multiple preoperative endoscopic ileal biopsies did not show the presence of inflammation or epithelioid granulomas. Preoperative endoscopic biopsies may not be helpful to exclude malignancy. Similar observations were seen in our case. Based on clinical and radiological findings, a neoplastic process was suspected. Resection of ileum, ileocaecal junction, appendix and part of transverse colon were done.

Classic microscopic pathologic appearance of xanthogranulomatous appendicitis show numerous lipid-laden histiocytes, abundant hemosiderin, multinucleated giant cells admixed with cholesterol clefts and mixed inflammatory infiltrate of polymorphs, lymphocytes, and plasma cells.[10] The absence of transmural involvement by epithelioid granulomas and Michaelis–Gutman bodies ruled out Chron's disease and malakoplakia, respectively.

Due to clinical and radiological resemblance of these lesions to neoplastic process, often warrant excision. They are often diagnosed postoperatively

CONCLUSION

Xanthogranulomatous appendicitis is a rare clinical entity. Clinical presentation is quite variable. Imaging modalities

and endoscopic findings alone are not adequate for definitive preoperative diagnosis. Clinical and radiological resemblances of these lesions to neoplastic process often warrant excision. Most cases are diagnosed after surgical resection.

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Conflicts of interest
There are no conflicts of interest.

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