ABSTRACT
Xanthogranulomatous reaction can occur in any organ but the most common sites are kidney and gallbladder. Xanthogranulomatous appendicitis (XA) is a rare clinical entity. There are a few case reports of XA diagnosed on histopathology but none on cytology. Here we report a case of a 47-year-old lady who presented with acute abdomen and was found to have a mass lesion in the right iliac fossa. She was diagnosed with XA intraoperatively on imprint cytology that was subsequently confirmed on histopathological examination. Due to the rarity of XA itself and the use of imprint cytology for intraoperative diagnosis the case is being presented.

Key words: Appendicitis; imprint cytology; xanthogranulomatous appendicitis (XA)
Introduction

Xanthogranulomatous inflammation is characterized by prominent histiocytic component with clusters of xanthoma type cells. It is regarded as an unusual healing pattern of appendicitis in contrast to the conventional pattern.[1] This type of tissue reaction can involve any organ but the most common sites are kidney and gallbladder.[2,3] Appendix is a very uncommon site for xanthogranulomatous reaction. Only a few case reports are available in the literature. Most of the patients present with a picture of acute or subacute abdominal pain and occasionally with a mass lesion in the right iliac fossa.[4] Mass lesions in the right iliac fossa can mimic locally advanced cancer but they have a benign course and can be cured by surgical resection. To avoid extensive resection, intraoperatively, imprint cytology can be used to decipher the cause of the mass. Due to the rarity of XA itself and the use of imprint cytology for intraoperative diagnosis the case is being presented.

Case Report

A 47-year-old female was brought to the surgical emergency room with the complaints of severe pain abdomen, vomiting, and fever. On clinical examination, there was tenderness on the McBurney’s point, rebound tenderness, and a palpable lump. Routine blood tests revealed a total leucocyte count (TLC) of $14.0 \times 10^9/L$ and the erythrocyte sedimentation rate (ESR) was 90 mm lst hour. She was conservatively managed for a day but the pain did not subside. Ultrasonography remained inconclusive and computed tomography (CT) scan of the abdomen showed a large soft-tissue mass in the right lateral pelvis with adherent loops of intestine. The walls of intestine were thickened along with three enlarged lymph nodes in the vicinity. Radiologically differential diagnosis of inflammatory versus neoplastic mass was given and the patient was taken up for exploratory laparotomy.

On exploration, a jumbled up mass in the right iliac fossa with adherent and thickened large intestine and lymph nodes was identified. Intraoperative imprint cytology was performed following which a limited right colon resection and lymph node removal were done.

Cytology

Smears revealed benign glandular epithelial cell groups and sheets of xanthoma cells along with multinucleate histiocytic giant cells in the background of neutrophils and mononuclear inflammatory cells [Figure 1].

Pathology

A jumbled up mass, comprising 2-cm part of small intestine, 5.5-cm long swollen appendix, adherent omentum, and 3-cm long large intestine, was received along with six lymph nodes.

Cut surface of the appendix showed markedly thickened wall with obliterated lumen showing gray white and yellow foci.

Microscopic examination revealed appendicular lining with focal ulceration and dense inflammatory cell infiltrate in the wall comprised of lymphocytes, plasma cells, histiocytes, eosinophils, foci of sheets of foamy macrophages, pools of extravasated mucin, marked foreign body giant cell reaction, and fibroblastic reaction against it [Figure 2]. Lymph nodes showed follicular hyperplasia.

Discussion

Xanthogranulomatous inflammation is a well-documented but uncommon entity that occurs at many sites in the
body. Though initially described in kidney in 1944, it occurs in other organs including fallopian tubes, endometrium, testicle, female and male genital tract, gallbladder, pituitary, colon, retroperitoneum, adrenal, and appendix.\(^3,5\)

Xanthogranulomatous appendicitis (XA) is a rare entity with only 11 cases reported in literature.\(^3\) Of these three cases presented as mass lesion, Chuang et al. in 2005 reported a case of a 39-year-old man who was admitted with fever, lower abdominal pain, and a mass in the right iliac fossa. With a suspicion of cancer, hemicolectomy was performed but on histopathological examination it turned out to be XA. The case illustrated that XA may mimic a locally invasive cancer.

Omar et al.,\(^6\) in 2011 reported a rare case of XA and cecal angiolipoma in a patient who was admitted with acute appendicitis and an appendicular mass.

In 2013, Mado et al.\(^7\) reported a case of a 78-year-old patient with abdominal pain, and the CT scan showed a 4-cm irregular mass near the caecum. The patient was operated with a clinical diagnosis of appendiceal mucocele, but the histopathological findings were consistent with XA.

Our case also presented with a mass lesion in the right iliac fossa and was explored with the possibility of carcinoma colon. Intraoperatively imprint smears were made that were suggestive of xanthogranulomatous inflammation. Findings were subsequently confirmed after the resection on histopathological examination.

The proposed mechanisms for the occurrence of xanthogranulomatous inflammation are defective lipid transport; immunological disorders altering the chemotaxis of leucocytes and macrophages; reaction to a specific infectious agent, such as \textit{Proteus} and \textit{Escherichia} species; infection by low virulence organisms; and lymphatic obstruction.\(^3\) The particular mechanism eliciting XA are organ obstruction, suppurative infection, hemorrhage, defective lipid transport, and local hypoxia.\(^6\) Histopathology of delayed (interval) appendectomy specimens shows higher incidence of XA.\(^7,8\)

Our case was remarkable for the finding of XA in a 47-year-old female who presented with acute abdomen and a mass lesion in the right iliac fossa that is suggestive of a cecal growth. This case is reported because of its rare presentation and the possibility that XA should be considered in the differential diagnosis of carcinoma colon. This case also shows the usefulness of on-site touch cytological evaluation for quick diagnosis.

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Conflicts of interest
There are no conflicts of interest.

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