CASE REPORT

Xanthogranulomatous Appendicitis: A Rare Case Report

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Abstract:

Xanthogranulomatous inflammation is a form of chronic inflammation where the exact eitiology is not known. It is commonly reported in organs like kidney and gall bladder Very few cases of xantho-granulomatous inflammation of appendix have been reported in the literature. We report a case of 30 year female presenting with pain abdomen and fever for which provisional diagnosis of appendicitis was made. Open appendicectomy was performed which on intraoperative examination showed that appendix was adherent to surrounding structures forming a mass. Resection of the mass was done and sent for histopathological examination. Gross examination of the received specimen revealed multiple irregular tissue bits and one tubular tissue bit. Multiple sections studied from resected tissue showed features of xanthogranulomatous inflammation in appendix and periappendicular tissue.

Keywords: Appendix, Periappendicular Tissue, Xanthogranulomatous Inflammation.

Introduction:

Xanthogranulomatous Inflammation (XI) is a well defined form of chronic inflammatory condition. It has been most commonly reported in organs such as the kidney and gall bladder. Very few cases of XI are reported in other organs like endometrium, epididymis, fallopian tubes, bone, skin, appendix, urinary bladder, thyroid and adrenal glands[1]. Acute appendicitis is one of the commonest surgical conditions; however Xantho-granulomatous Appendicitis (XA) is a rare finding in resected specimens of acute appendicitis. The literature search revealed only few case reports of XA [2].

Case Report:

A 30 year female patient presented to our hospital with complaints of pain in the right iliac fossa for the past 3 weeks which was insidious in onset and gradually progressive in nature which aggravated on doing physical work. The pain was non-radiating and was relieved on taking medications. Patient also had an episode of fever two days back which was of moderate grade and not associated with chills or rigor. Patient had no history of vomiting, bowel disturbance, burning micturition and respiratory complaints. She had been a tobacco chewer since 10 year of age. There was no history suggestive of tuberculosis, diabetes mellitus, hypertension or myocardial infarction. Haematological and biochemical parameters were within the normal range.

On general physical examination pallor was present and scaling of skin over arms and neck was also present. On palpation tenderness was elicited in the right iliac fossa and right lumbar region. There was no guarding or rigidity on palpation and there was no lymphadenopathy. On the basis of history and examination provisional diagnosis of appendicitis was made and open appendicectomy was planned.

After opening abdomen, appendix was found to be inflammed and densely adherent to surrounding

structure forming a mass. The appendix was relieved from all its adhesions, mesoappendix was ligated and then the appendix was excised and sent for histopathological examination.

Histopathology:

Gross:

The laboratory received multiple irregular tissue pieces with the largest measuring 4x2 cm and smaller measuring 3.5x1 cm and one tubular tissue bit measuring 1.5 cm (Fig.1). The cut surface of the irregular tissue pieces showed solid light brown appearance with focal yellowish areas (Fig. 1 Inset). The cut surface of tubular structure showed narrowing of the lumen.

Microscopy:

Section studied from the appendix showed extensive mucosal ulceration. The base of the ulcer showed granulation tissue with dense and diffuse mixed inflammatory cell infiltrate with prominent cells being foamy histocytes arranged in sheets, clusters and scattered singly (Fig.2 and Fig. 3). Also it was noted, few ill formed granulomas and foreign body giant cells (Fig.4). Meso-appendix also showed granulation tissue with foreign body reaction. A Ziehl Neelsen stain for acid fast bacilli was negative.

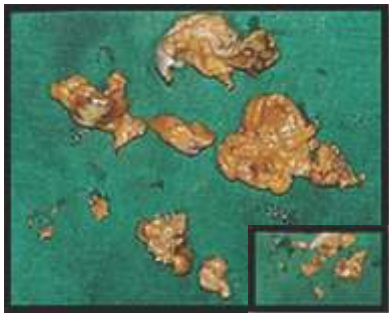

Fig. 1: Gross Photograph showing Irregular Tissues Bits and a Tubular Tissue Piece. Inset-Focal Yellowish Area on Cut Surface

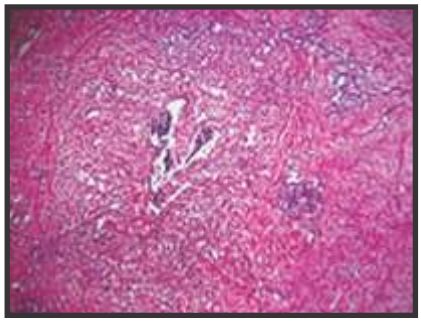

Fig. 2: Photomicrograph showing Sheets of Foamy Macrophages in the Wall and Periappendicular Tissue (H&E100X)

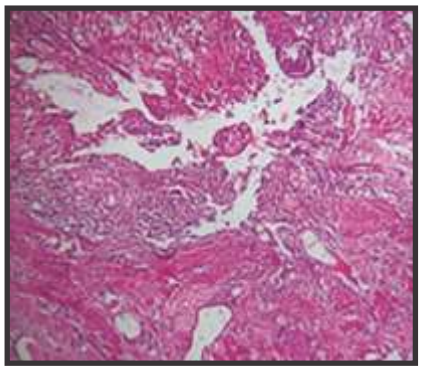

Fig. 3: Photomicrograph showing Sheets of Foamy Macrophages in the Wall and Periappendicular Tissue (H&E400X)

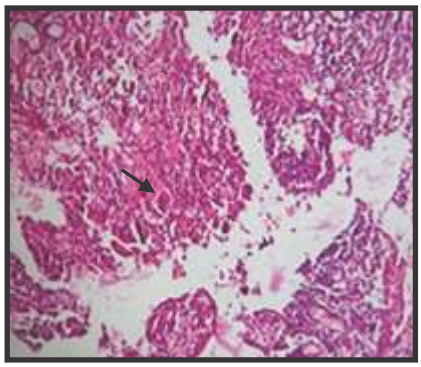

Fig. 4: Arrow showing Multinucleate Giant Cells in the Wall of Appendix (H&E 400X)

Discussion:

XI is a pathological entity having unique morphological features which includes golden yellow mass like lesion associated with microabscesses and large number of foamy histocytes. XA should be differentiated from other lesions containing foam cells like malakoplakia and localized xanthoma. Malakoplakia shows characteristic presence of Michaelis Gutmann bodies with inflammatory reaction in the affected tissue. However in the localized Xanthoma there is no parenchymal destruction and only aggregates of foam cells without inflammatory reaction [2]. In the present case, appendix and periappendicular tissue showed features of tissue destruction.

Sometimes XI of appendix can masquerades as malignancy clinically due to presence of a diffuse inflammatory and fibrotic changes and the presentation of a mass like lesion and hence clinically can be misdiagnosed as malignant lesion [2].

Omer *et al*. [3] mentioned that granulomatous inflammation in appendix can be noted in ruptured acute appendicitis. In the case reported by Omer *et al*. patient was treated with antibiotic therapy inspite of which patient presented with rupture of appendix and it was postulated that ruptured appendix might be the cause of XI in appendix [3]. In the present case histopathological study of appendix showed features of appendicular perforation with periappendicitis. A similar explanation may hold for XI in the present case.

It was also reported by some authors that, delayed or interval appendicectomy specimens often show an inflammatory reaction with granulomas, XI, mural fibrosis thickening, and transmural chronic inflammation. These changes can mimic Crohn's disease microscopically. Hence, Crohn's disease in the small or large bowel should be ruled out in patients of XA [3].

According to some authors several factors may precipitate XA. These factors were organ obstruction, suppurative inflammation; hemorrhage and local hypoxia. These authors excluded possibility of single pathophysiology of XA as a spectrum of various appendicular lesions were noted in XA [4]. In some studies it was mentioned that XA probably represents a chronic inflammatory process leading to tissue destruction and localized proliferation of macrophages containing large amounts of lipid [5]. A literature review of various case reports of XA revealed that deficiency in lipid transport, disturbance of leucocyte, macrophage chemotaxis, lymphatic obstruction and infection of Proteus and Escherichia species may be precipitating factors for XA [2, 6].

Conclusion:

XI of appendix is a rare form of chronic inflammation. Precipitating factors for XI of the appendix are exactly not known. Various authors put forth various hypothesis for etiology of XA. Hence awareness of this lesion with underlying precipitating factor can help in proper management of the patient by treating the underlying cause.

References

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