

Case Report

Xanthogranulomatous appendicitis an incidental finding

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Abstract: Xanthogranulomatous inflammation is a clinical finding which has been reported in various locations, such as kidney. Although appendicitis is common xanthogranulomatous appendicitis rare.

Here we describe a twenty nine year old woman with history of cesarean section that developed abdominal pain in right lower quadrant. With impression of appendicitis this patient was operated. Pathologic specimen was reserved and Xanthogranulomatous was approved with H&E and IHC staining.

Key word : *Xanthogranulomatous , appendicitis, Immunohistochemistry*

1. Introduction

Xanthogranulomatous inflammation is an uncommon, though well-recognized entity that has been described in various organs but mostly commonly in the kidney and gallbladder (1).

This inflammation characterized histologically by presence of high number of foamy histiocytes admixed with lymphocytes and plasma cells.(2)

Although acute appendicitis is a very common surgical condition, xanthogranulomatous appendicitis is a rare phenomenon.(3)

2. Case

A 29 years old woman with history of cesarean section about 3years ago, developed abdominal pain in right lower quadrant for 2 month. on evaluation she was found to have tenderness in the McBurney's point along with rebound tenderness. her ultrasonogram was inconclusive. Routh blood test show a white blood count 13000 cell/microliter with 80% neutrophils, hemoglobin 13 gr/dl, platelet 458000 cell/microliter as well as increased ESR level (30 mm/hours). other results were normal.

Then she was clinically diagnosed as a case of acute appendicitis and exploratory laparotomy was performed.

Per operatively the appendix appeared hyperemic. No gangrenous change or perforation was noted.

3. Pathologic Findings:

We received the specimen of appendix in formalin, which measured 7.5 cm in length and 1cm in maximum diameter. The external surface appeared congested and dull. Cut surface showed congested mucosa with yellow colored areas. The lumen containing fecaloid material.

The Hematoxylin and Eosin (H&E) stained sections from the appendiceal wall reveal focal neutrophilic infiltration in mucosal surface as well as muscularis layer. Large areas of the muscular and serosal layers were replaced by collections of foamy histiocytes as well as foreign body-type multinucleated giant cells, lymphocytes neutrophils and congested blood vessels.(figure .1A-1B)

Special stains were done. No acid-fast bacilli were seen on Ziehl-Neelson stain. Von Kossa (for calcium) and Perl's stain (for iron) were done to rule out presence of Michaelis Gutmann bodies, which shows positivity for both iron and calcium stains.

Immunohistochemistry (IHC) was done with CD 68 which showed strong positivity in the foamy cells (Fig. 2)

These cells were negative for pan cytokeratin(CK) (Fig. 3).

Finally based on H&E, special stains and IHC findings, a diagnosis of xanthogranulomatous appendicitis was offered. Post-operative period was uneventful and presently the patient is asymptomatic.

4. Discussion

Xanthogranulomatous inflammation is a rare form of chronic inflammation, manifested by the presence of

nodular or diffuse collection of granular, eosinophilic, PAS positive histiocytes, lipid-laden macrophages admixed with lymphocytes, plasma cells, neutrophils, and often multinucleated giant cells with or without cholesterol clefts. Occasionally, granulation tissue, and necrotic debris are observed with reactive lymphoid hyperplasia. (3,4)

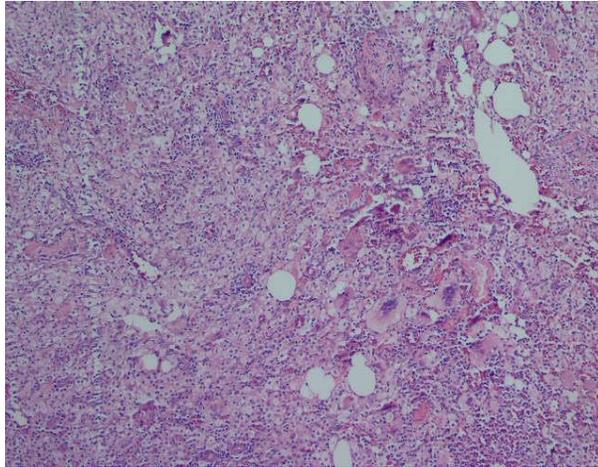


Fig. 1-A: a cut of large areas of the muscular and serosal layers

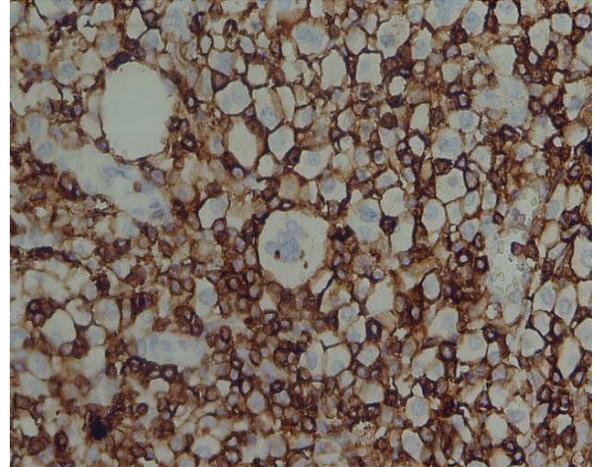


Fig. 2: Immunohistochemistry (IHC) with CD 68

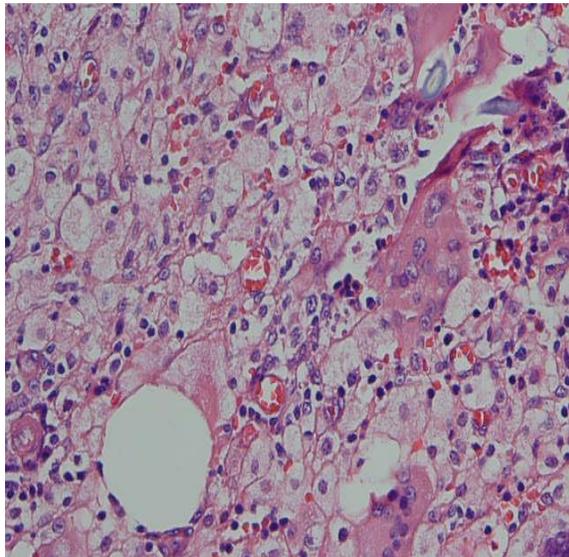


Fig. 1-B: a cut of large areas of the muscular and serosal layers

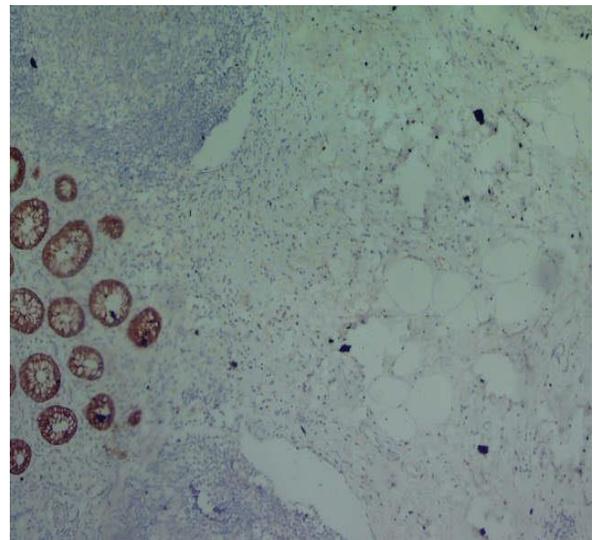


Fig. 3: pan cytokeratin positive in appendiceal mucosa/negative in histiocyte-like cells

The exact etiology of xanthogranulomatous inflammation is uncertain. Proposed etiologies include defective lipid transport, immunologic disturbances a specific immune response toward Proteus and Escherichia infections, and lymphatic obstruction (3,5).

Other lesions containing foam cells should be distinguished from XGI. Malacoplakia is characterized by an inflammatory and destructive xanthomatous proliferation with the presence of Michaelis–Gutmann bodies or calco-spherites Small localized xanthoma deposits without parenchymal destruction or xanthomas with prominent foam cell features must also be considered in the differential diagnosis(6). Some time it is not possible to differentiate XGI from an infiltrative cancer because XGI might present as a mass-like lesion with an extension of fibrosis and inflammation to the surrounding tissues, mimicking an infiltrative cancer. (7).

5. Conclusion

To conclude, the aim of presenting this case was to highlight the rarity of XGI of the appendix, presenting as an acute appendicitis

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