Case Series

Encountering Xanthogranulomatous Inflammation in a Myriad of Settings

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Background: Xanthogranulomatous inflammation is a rare but well recognized variety of chronic inflammation seen in multiple sites. It may present as a mass lesion and mimic malignant tumors, in other cases it may be discovered as an incidental finding in tissues removed for different reasons. Xanthogranulomatous inflammation was encountered in five different cases which included two gall bladders, one kidney, one endoscopic biopsy from gastric mucosa and one collaural fistula. The demographic data as well as pathological findings are described. After Although the exact etiopathogenesis of xanthogranulomatous inflammation is still debated, it is agreed that it has no pre-malignant potential. Therefore, resection is considered adequate therapy for effected patients. This makes it all the more important to recognize it in its various morphological forms, even in endoscopic biopsies, to avoid unnecessary extirpative procedures.

Key words: Xanthogranulomatous inflammation, foamy macrophages, collaural fistula, endoscopic biopsy.

Introduction

Xanthogranulomatous inflammation is a rare but well recognized variety of chronic inflammation. [1]It is described most often in kidney, gall bladder, appendix and colon, but involvement of female genital tract, breast and branchial cleft cyst have also been reported.[2]It may present as a mass lesion and mimic malignant tumors. Its etiopathogenesis remains obscure though infection, outflow obstruction, abscess formation and hemorrhage have been advanced as possible incriminants by different authors.[1-3] Recognition of the entity in its various forms is important because, as stated earlier, it could be mistaken for a malignant tumor resulting in unnecessary resective surgeries.[4,5] It was seen in five of our patients recently and the findings are being summed up in this paper.

Materials and Methods

This paper deals with five cases diagnosed with xanthogranulomatous inflammation. There were two gall bladders, one kidney, one endoscopic biopsy from gastric mucosa and one collaural fistula. All specimens were received in Pathology Department of PGMI, Lahore from Lahore General Hospital, Lahore. One of the gall bladders and the gastric biopsy had been submitted with the suspicion of a malignant growth. The demographic details as well as findings are summarized in Table. All cases were diagnosed on hematoxylin and eosin stained slides. PAS stain was employed where indicated. The microscopic findings are depicted in Fig 1-3.

All cases contained collections of foamy macrophages (Fig 1-3) with a variable admixture of giant cells, lymphocytes and plasma cells. These

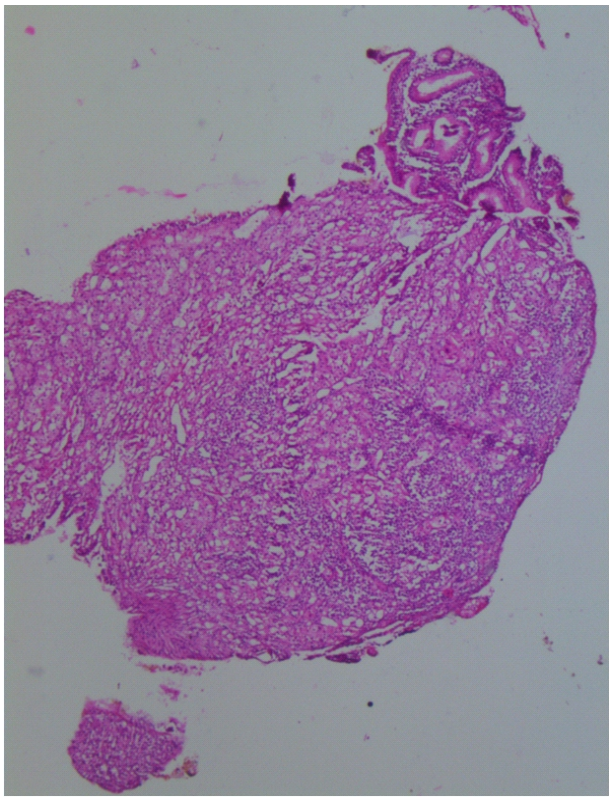

Fig-1: Photomicrograph showing endoscopic biopsy from gastric mucosa. There are sheets of foamy macrophages admixed with lymphocytes and plasma cells. (H &E, x40)

Collections replaced the native architecture to a variable degree. Cases with a short clinical history comprised mostly of rounded histeocytes with foamy to granular cytoplasm (Fig 1,2). Cases with a prolonged history showed similar cells admixed with spindle shaped cells (Fig 3). In these cases the granulomatous nature of the lesions was more obvious. They also showed adherence to and involvement of surrounding structures. The kidney and one of the gall bladders showed these changes.

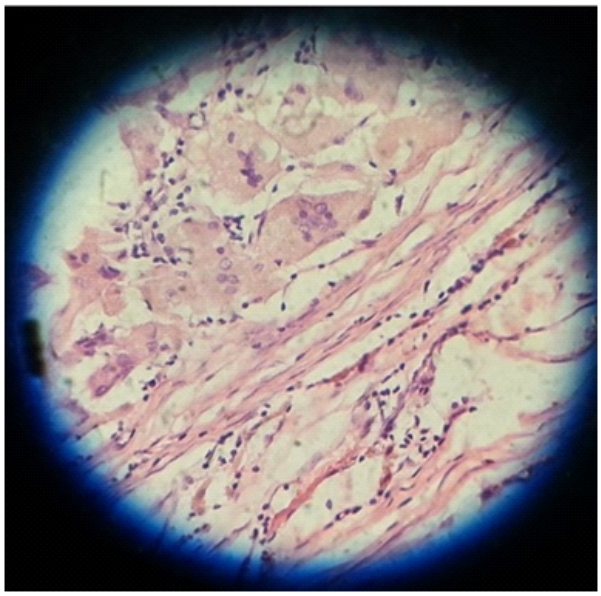

Fig-2: Photomicrograph showing the wall of collaural fistula. The lining epithelium is replaced by foamy macrophages, histeocytes with granular cytoplasm, lymphocytes, plasma cells and macrophages. (H & E, x 400)

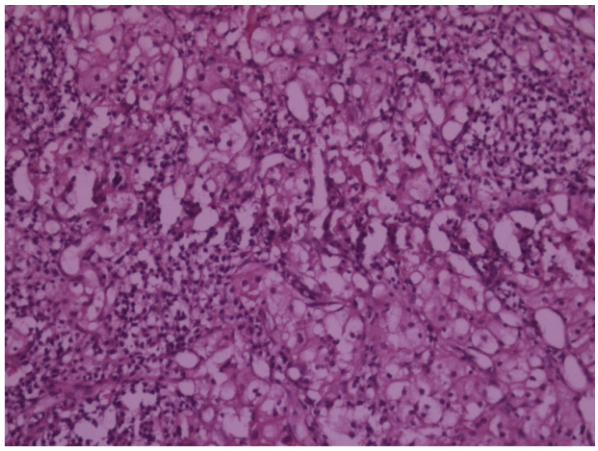

Fig-3: Photomicrograph showing xanthogranulomatous inflammation in kidney. There is an admixture of foamy macrophages with spindloid cells, lymphocytes and plasma cells. (H & E, x 100)

Discussion

Xanthogranulomatous inflammation was first described by Schlagenhaufer in 1916.[6] Since then it has been reported in many different organs and tissues. Its importance stems from its ability to form a mass, a fact borne out by terms like 'inflammatory tumor of the kidney". It has no pre malignant potential.[7]

The exact cause and pathogenesis of the entity have not been established so far. As stated above numerous factors have been considered. Of these infection and outflow obstruction are said to be the most important, though the role of mucosal breakage and leakage of contents into deeper tissues has also found favor.[1-3,8] Several authors have described the temporal

Table-1: Table showing the demographic data and morphological findings of cases..

Organ/ Tissue	Age and Clinical gender of diagnosis the patient	Clinical Diagnosis	Gross Findings	Microscopic Ffindings
Gallbladder	35yr, M	Chronic cholecystitis	Thickened, ulcerated wall with yellow spots	Sheets of foamy macrophages admixed with lymphocytes, plasma cells and areas of hemorrhage. Cholesterol clefts.
Gallbladder	42yr, M	Adenocarcinoma gallbladder	Thickened, ulcerated wall with yellow spots	Foamy macrophages, spindle shaped cells, admixed with lymphocytes and plasma cells. Numerous micro abscesses and areas of hemorrhage.
Kidney	54yr, M	Chronic pyelonephritis	Scarred kidney with multiple stones	Micro abscesses, areas of hemorrhage, spindle shaped histeocytes, fibrosis, cholesterol clefts
Gastric mucosa	60yr, M	Carcinoma Stomach	Routine endoscopic biopsy	Sheets of foamy macrophages with lymphocytes and plasma cells.
Collaural Fistula	35yr, M	Collaural fistula	Multiple grey white soft tissue pieces	Foamy macrophages replacing the lining epithelium, occasional giant cells.

changes seen in cases of xanthogranulomatous inflammation. These include the gradual replacement of rounded foamy and granular histeocytes by elongated, spindloid cells.[3,9] This metamorphosis was seen in our cases too; those with a longer duration had a greater proportion of elongated cells and fibrosis **(Table, Fig 1-3).**

Another notable feature is the widespread tissue destruction seen around areas of xanthogranulomatous inflammation. Most authors agree that the former predates the latter, i.e., first there is tissue destruction then there is replacement by the specific components associated with xanthogranulomatous inflammation. A small subset of workers is of the opinion that the process may be self-perpetuating and once begun may continue to smolder and spread. In either case, what is undebatable is the destruction and dissolution of surrounding parenchyma and its eventual replacement by fibrous tissue.[3,10,11] This leads to adherence to neighboring organs, difficulty in surgical excision and increased suspicion of a malignant nature **(Fig 3).** Cases are on record where xanthogranulomatous inflammation was over-diagnosed as a malignant tumor resulting in unwarranted resections.[3,5,8,12]

This paper is being written to highlight the fact that possibility of this lesion should be entertained in the differential diagnosis of masses from a variety of described, as well as yet undescribed, tissue sites. Since it is an inflammatory condition and has no pre-malignant potential, resection is considered adequate therapy for effected patients.[2,8]

Conclusion

Although the exact etiopathogenesis of xanthogranulomatous inflammation is still debated, it is agreed that it has no pre-malignant potential. Therefore, resection is considered adequate therapy for effected patients. This makes it all the more important to recognize it in its various morphological forms, even in endoscopic biopsies, to avoid unnecessary extirpative procedures.

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