CASE REPORT

Xanthogranulomatous Inflammation of the Maxilla Mimicking a Dentigerous Cyst: A Rare Case Report

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ABSTRACT

Xanthogranulomatous inflammation is a chronic inflammatory condition rarely encountered in the head and neck region. We presented here Xanthogranulomatous inflammation involving the maxilla in a 33 -year -old male.

Key words: Xanthogranulomatous inflammation, chronic inflammation, maxilla.

INTRODUCTION

Xanthogranulomatous inflammation (XGI) is a rare form of chronic inflammatory response characterized by lipid-laden foamy macrophages, multinucleated giant cells, and fibrosis.[6,7] It most commonly involves the kidney and gallbladder, while involvement of the head and neck region is exceedingly rare and largely confined to isolated case reports and small clinicopathological series.[3,6] In a clinicopathological series of 31 cases, involvement of the oral cavity accounted for only 6.4% of xanthogranulomatous inflammatory lesions, underscoring the rarity of head and neck involvement.[7]

In the maxillofacial region, XGI is uncommon and may clinically and radiologically mimic odontogenic cysts or benign tumours, resulting in significant diagnostic difficulty.[1,3] The etiopathogenesis of XGI remains poorly understood; however, chronic inflammation, prior trauma, foreign body reaction, and ineffective clearance of inflammatory debris have been implicated as possible contributing factors.[6,7] Imaging and cytological findings are often non-specific, making histopathological examination supported by immunohistochemistry essential for establishing a definitive diagnosis.[6,7] We report a rare case of xanthogranulomatous inflammation of the maxilla with extension into the maxillary sinus, initially suspected to be a dentigerous cyst.

CASE PRESENTATION

A 33-year-old male presented with a painless, progressively enlarging swelling over the left cheek for six months, with no history of fever, dental infection, or difficulty in mastication. He had a significant past history of facial trauma, for which surgical fixation of the left maxilla using an iliac crest bone graft had been performed several years earlier. Fine-needle aspiration cytology suggested a cystic lesion. Non-contrast computed tomography of the face revealed a well-defined hypodense lesion measuring 3.7 × 3.6 × 3.7 cm arising from the upper alveolar ridge of the left maxilla with extension into the maxillary sinus, causing bony expansion and remodelling, radiologically suggestive of a dentigerous cyst. The lesion was excised via a sublabial approach; intraoperatively, it was poorly circumscribed and lacked a well-defined capsule. Histopathological examination showed fibrocollagenous tissue with marked xanthomatous reaction, numerous foamy histiocytes, and multinucleated giant cells. Immunohistochemistry demonstrated CD68 positivity and calretinin negativity, confirming xanthogranulomatous inflammation. The postoperative period was uneventful, and

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at 2-month follow-up the patient remained asymptomatic with no evidence of recurrence.

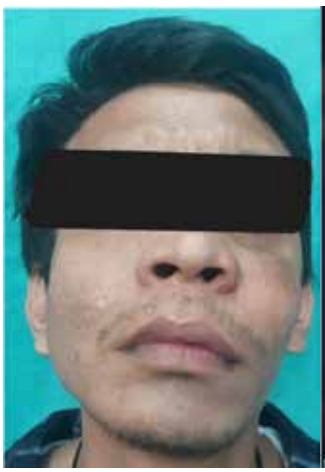

Fig 1: Post Operative picture showing reduced swelling

Fig 2: Dimensional Computed Tomography scan showing lesion on left maxilla

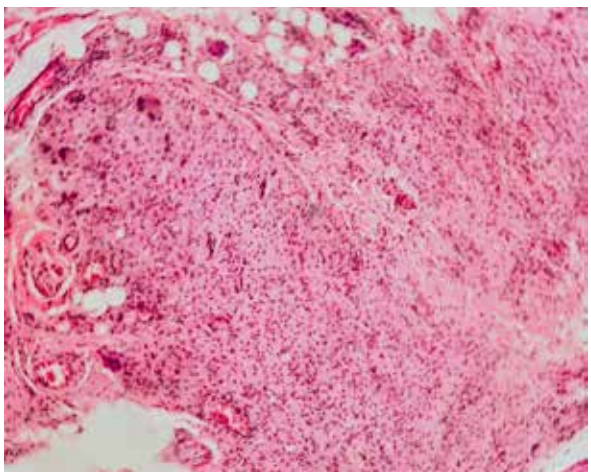

Fig 3: Histopathology showing sheets of foamy histiocytes along with a few scattered multinucleated giant cells suggestive of xanthogranulomatous inflammation

DISCUSSION

Xanthogranulomatous inflammation is a rare pathological entity in the maxillofacial region and has been reported only sporadically in the head and neck.[1,3] It predominantly affects the kidney and gallbladder, while involvement of the head and neck region remains exceedingly uncommon.[3,6] The exact etiopathogenesis of XGI remains unclear; however, several mechanisms have been proposed, including chronic inflammation, persistent infection, lipid metabolism abnormalities, haemorrhage, obstruction, and foreign body reaction.[6,7] In the present case, the history of prior facial trauma with surgical fixation of the left maxilla using an iliac crest bone graft may have acted as a predisposing factor by inducing chronic inflammation or a foreign body–related response.

Clinically and radiologically, XGI often mimics odontogenic cysts such as dentigerous or radicular cysts, as well as other benign tumours, making preoperative diagnosis challenging.[1,3] Imaging findings are usually non-specific and may demonstrate cystic lesions with bony expansion, as observed in the present case where radiological features strongly suggested a dentigerous cyst.[1] Fine-needle aspiration cytology may also be misleading or inconclusive, further limiting the ability to establish an accurate preoperative diagnosis.

Definitive diagnosis relies on histopathological examination supported by immunohistochemistry.[6,7] The presence of lipid-laden foamy histiocytes and multinucleated giant cells, highlighted by CD68 positivity, confirms the histiocytic nature of the lesion, while negative calretinin staining helps exclude granular cell tumour and other important differential diagnoses.[6] The absence of a well-defined capsule noted intraoperatively in the present case further supports the inflammatory rather than true cystic nature of the lesion.

Complete surgical excision is considered the treatment of choice for xanthogranulomatous inflammation, as the lesion represents a chronic destructive inflammatory process that does not regress spontaneously.[1,6] If left untreated, XGI may progressively enlarge, cause local bone destruction, and continue to mimic neoplastic conditions, potentially leading to functional impairment and diagnostic confusion.[3,6] Surgical excision not only provides definitive diagnosis but also prevents further local tissue damage and recurrence, with reported outcomes being favourable.[1,5]

There is no established role for medical management alone in xanthogranulomatous inflammation. Antibiotics or anti-inflammatory therapy may provide temporary symptomatic relief but are insufficient to eradicate the lesion due to its chronic granulomatous nature.[6,7] Although no standardized follow-up protocol exists, periodic clinical follow-up, with radiological evaluation when indicated, is recommended to detect early recurrence, particularly in the early postoperative period.[1,5]

CONCLUSION

Xanthogranulomatous inflammation of the maxilla is an exceptionally rare inflammatory condition that can closely mimic odontogenic cysts and other benign lesions both clinically and radiologically, thereby posing a significant diagnostic challenge.[1,3] This case highlights the importance of maintaining a high index of suspicion for xanthogranulomatous inflammation in the differential diagnosis of cystic lesions of the maxilla, particularly in patients with a history of trauma or prior surgical intervention.[1,6]

Definitive diagnosis relies on thorough histopathological examination supported by immunohistochemistry, which is essential to distinguish this entity from neoplastic and other

inflammatory conditions.[6,7] Recognition of the characteristic pathological features helps avoid misdiagnosis and unnecessary aggressive treatment. Complete surgical excision remains the treatment of choice and provides both diagnostic confirmation and definitive management, with favourable outcomes and a low risk of recurrence on follow-up.[1,5]

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