

## Case Report

# Xanthogranuloma of nose in an adult: an uncommon presentation

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### ABSTRACT

Xanthogranuloma is a relatively rare histiocytic cutaneous disorder that typically affects the pediatric age group. Its occurrence in adulthood is still rarer with only a handful of cases on the record. The present case report describes a case of adult-onset xanthogranuloma in a 49 years old lady who presented to the otorhinolaryngology clinic with a 6 months history of a nodule on the left side of the columella. There was no history of associated pain or bleeding from the site. Local examination revealed a 0.6×0.6 cm, non-tender, soft-to-firm, pinkish dome-shaped lesion with rounded edges. No similar lesions were found elsewhere. Excisional biopsy of the lesion was performed and microscopy showed a dermal lesion comprising of sheets of histiocytes admixed with lymphoplasmacytic infiltrate, touton giant cells and few spindle cells. A diagnosis of xanthogranuloma was rendered. A number of lympho-histiocytic lesions comes in the differential diagnosis of xanthogranuloma and hence it is important to be aware of such unusual presentations in order to make correct histological diagnosis.

**Keywords:** Xanthogranuloma, Adult, Nose, Touton giant cells

### INTRODUCTION

Xanthogranuloma is an uncommon non-langerhans histiocytic cutaneous lesion of unknown etiology.<sup>1</sup> Although typically encountered in children and adolescents, about 10% of the cases can occur during adulthood.<sup>1</sup> Adult onset xanthogranuloma (AXG) of nose is a rare phenomenon with only a handful of cases recorded in the literature. Here, we reported another such rare case of AXG involving the nasal columella.

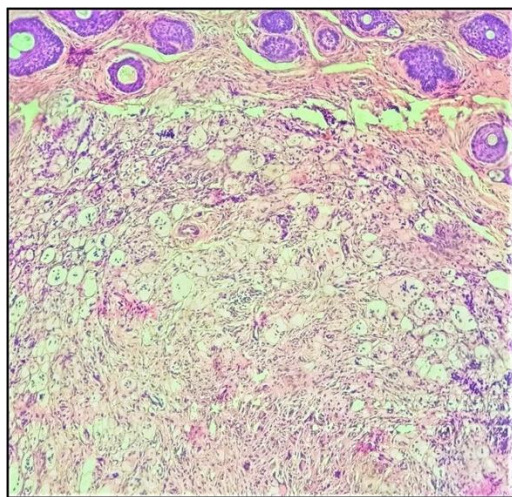
### CASE REPORT

A 49-years-old lady presented to the otorhinolaryngology outpatient clinic with a 6-months history of a gradually progressive nodule on the left side of the columella. There was no history of associated pain or bleeding from the site or prior trauma to the site. There was no significant past medical or family history either. Local examination revealed a 0.6×0.6 cm, non-tender, soft-to-firm, pinkish dome-shaped lesion with rounded edges. No similar

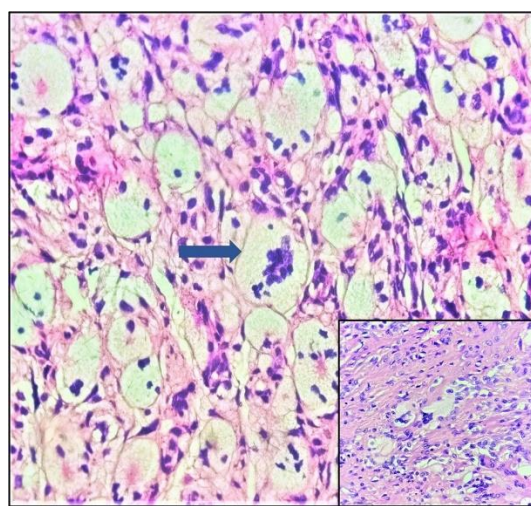
lesions were found elsewhere. No additional lesions were detected on flexible nasal endoscopy. Remaining head and neck examination and systemic examination were unremarkable.

The routine laboratory test results (complete blood counts, coagulation profile, renal and liver function tests, random blood sugar levels, lipid profile and urine analysis) were also within normal limits. Excisional biopsy of the lesion was performed and the histopathological picture was diagnostic of xanthogranuloma. Microscopy showed a dermal infiltrate comprising of sheets of foamy histiocytes admixed with lymphoplasmacytic infiltrate, touton giant cells and few spindle cells (Figure 1 and 2).

No areas of necrosis/increased mitosis/atypia were noted. Special stains, including Periodic acid Schiff (PAS), Wade-Fite (WF), Giemsa and Gram stain were negative for microbial agents. The overlying epidermis was largely unremarkable. At six-month follow-up, there was no evidence of recurrence.



**Figure 1: Sheets of foamy histiocytes in the dermis (Hematoxylin and Eosin stain, at 40X).**



**Figure 2: Foamy histiocytes with scattered Touton type giant cells and lymphocytes. Inset shows focal areas of fibrosis (Hematoxylin and Eosin stain, at 400X).**

## DISCUSSION

Xanthogranuloma is a relatively rare histiocytic cutaneous disorder that typically affects the pediatric age group and is therefore, synonymously designated as ‘Juvenile xanthogranuloma’(JXA). JXG and AXG are almost identical in terms of site predilection, clinical appearance, microscopic picture and immunohistochemical profile. Both JXG and AXG are histologically characterized by dermal proliferation of histiocytes admixed with few giant cells and variable degree of inflammatory cell infiltrate. The histiocytes bear a xanthomatous appearance with foamy cytoplasm. The giant cells are of Touton type, although foreign body type of giant cells may also be present.<sup>2</sup> Early lesions may lack these giant cells.<sup>3</sup> Immunohistochemically, the lesional histiocytes are positive for CD68 and negative for S100, CD1a and

CD207, thereby differentiating it from Langerhans’ cell histiocytosis. In contrast to its juvenile form, AXG tends to persist more often than not.<sup>1,3,4,5</sup>

Gedela et al and Tan et al reported AXG of nasal columella and nasal ala respectively.<sup>2,6</sup> Similar to our case, these patients presented with slow-growing nasal nodule without any history of nasal obstruction or similar lesion elsewhere. Worden et al described AXG involving the pyriform aperture of the nose in a 73-year-old lady who presented with nasal obstruction.<sup>4</sup> She also had multiple yellow plaques over infraorbital area, shoulders, back and thighs.<sup>4</sup> Bastianpillai and colleagues reported a case of AXG of the right middle turbinate and uncinate process, which clinically mimicked as malignancy.<sup>7</sup> The patient presented with nasal obstruction and bloody discharge. Imaging showed the lesion involving the paranasal sinuses and extending superiorly up to the cribriform plate. Piecemeal tumor excision was performed and the histopathological examination established the diagnosis.<sup>7</sup> AXG of nose may pose diagnostic challenge to both surgeons and pathologists and other common lymphohistiocytic lesions of nose needs to be excluded. The differential diagnoses include rhinoscleroma, lepromatous leprosy, verruciform xanthoma and necrobiotic xanthogranuloma.<sup>3,4,8</sup> Hence, surgical excision with histopathological evaluation forms the standard mode of treatment as well as diagnosis. Special stains (PAS, WF, Giemsa and Gram stain) performed in this case, ruled out an infective etiology.

## CONCLUSION

AXG can rarely affect the nasal cavity and it is important to be aware of such unusual presentations in order to make a correct diagnosis. A number of lymphohistiocytic lesions comes in the differential diagnosis of xanthogranuloma and hence excisional biopsy with subsequent detailed histopathological analysis, accompanied by special stains and immunohistochemistry (if required), is mandatory. Also, the patient should undergo a thorough clinical evaluation and imaging to rule out the presence of similar lesion elsewhere.

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