Central xanthoma of the mandible in a 12-year-old boy

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BACKGROUND

Xanthomas are benign proliferations of lipid-laden macrophages that most often affect the skin and extensor tendons. The majority occur in the setting of systemic diseases associated with altered lipid metabolism, such as hyperlipidemia and diabetes mellitus. Intraosseous xanthomas are uncommon and can affect both the axial and appendicular skeleton.

CASE REPORT

Clinical and radiographic findings

A 12-year-old boy with a non-contributory medical history presented for evaluation of an asymptomatic radiolucency of the right mandible. Intraoral examination revealed no mucosal abnormalities or swelling in the region. Cone beam computed tomography (CBCT) imaging showed a well-defined, unilocular radiolucency associated with a mesioangular impacted #32 **(Fig. 1a).** Slight expansion of the buccal and lingual cortices was evident; however, no cortical perforation was observed **(Fig. 1b).** The lesion was completely enucleated in conjunction with extraction of #17 and #32.

Microscopic features

Histopathologic examination showed a proliferation of large cells with foamy to granular cytoplasm, varying in distribution from loose sheets to compact clusters with minimal intervening connective tissue **(Figs. 2a and b).** On immunohistochemical analysis, the lesional cells were strongly reactive for CD68 **(Fig. 2c),** and negative for S-100 **(Fig. 2d),** favoring a non-Langerhans cell histiocytic process. A diagnosis of central xanthoma was rendered.

Follow-up

A subsequent medical evaluation was found to be unremarkable for lipid abnormalities, endocrine disorders, and lipidoses. Follow-up CBCTs taken every 6-month demonstrated progressive bone fill **(Figs. 1c and d).** Unfortunately, new lytic areas were noted at the patient's most recent 15-month reevaluation. Re-excision is planned.

Radiographic findings

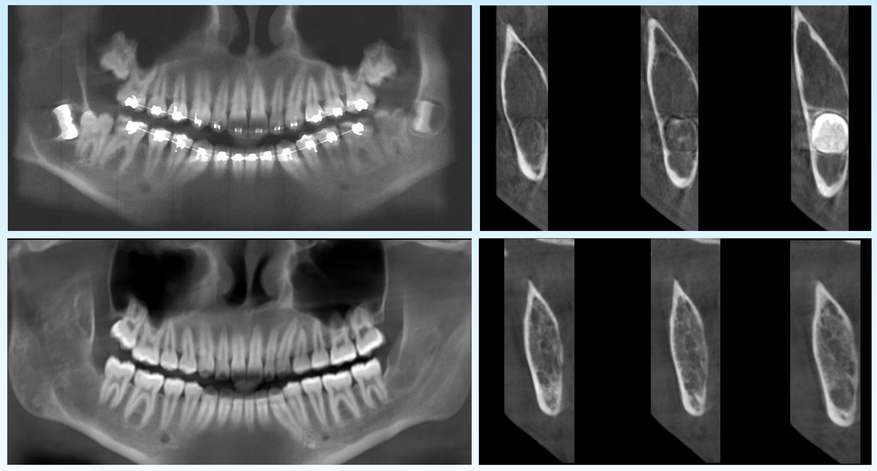

Figure 1. Unilocular radiolucency with corticated margins surrounding impacted #32. **A,B.** Initial presentation (panoramic, coronal slices; CBCT). **C,D.** One year post-operatively (panoramic, sagittal slices; CBCT).

Microscopic features

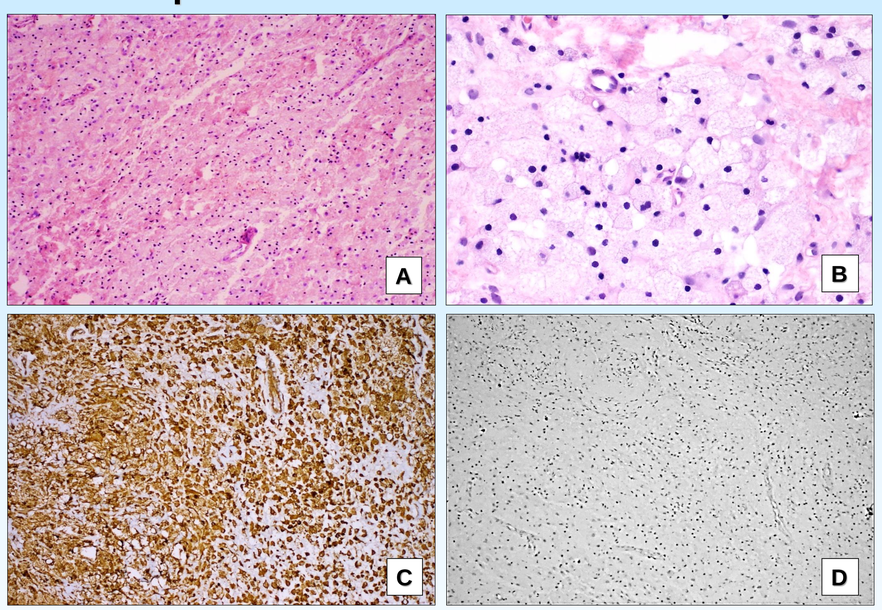

Figure 2. Microscopic images of the right mandibular biopsy. **A.** Low-power view showing syncytial sheets of large cells with eosinophilic cytoplasm (H&E, 10x); **B.** High-power view depicting cells with abundant foamy to granular cytoplasm and centrally-placed nuclei (H&E, 40x); **C.** Strong cytoplasmic staining with CD68 (20x); **D.** Negative staining with S-100 (20x).

DISCUSSION

- Gnathic xanthomas are exceedingly rare with less than 40 cases reported to date. Most arise in the second and third decades of life. A predilection for the posterior mandible is observed.
- The majority of gnathic lesions are asymptomatic. Pain and expansion are rarely described.
- The most common radiographic presentation is a unilocular radiolucency with "punched-out" or corticated margins. Association with an unerupted tooth is an unusual finding.
- Microscopic hallmarks include loose to compact sheets of histiocytic cells characterized by foamy or granular cytoplasm. Intervening stroma and bony trabeculae are typically scarce. The lesional cells exhibit a CD68+/S-100 negative immunoprofile.
- Diagnostic challenges can arise due to the nonspecific clinical and radiographic presentation. Microscopic overlap with other histiocytic disease processes can further confound diagnosis, making clinicoradiographic correlation necessary.
- Treatment of gnathic xanthomas consists of thorough curettage and recurrences are distinctly uncommon, especially in the absence of an underlying lipid dysregulation.
- Clinical presentation that precedes the diagnosis of a dyslipidemia or lipidoses may serve as a harbinger of underlying disease.

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