

Case Report

Juvenile cemento-ossifying fibroma: a case report using modified Gallego's staining

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Received: 12 December 2015

Accepted: 06 January 2016

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ABSTRACT

Juvenile cemento-ossifying fibroma is a rare benign but locally aggressive tumour with high recurrent potentials. The present study reports a case of juvenile aggressive ossifying fibroma involving the maxilla of a 6-year-old boy reported with a chief complaint of painless swelling over right side of the cheek since six months without any significant etiological factor. Modified Gallego's staining had been used to differentiate hard tissue deposits in the lesional tissues which have not been implicated so far in any fibroosseous lesions which makes this report unique.

Keywords: Juvenile cemento ossifying fibroma, Maxilla, Modified Gallego's stain

INTRODUCTION

Cemento-ossifying fibroma is a fibro-osseous lesion that arises from the periodontal membrane, which contains pluripotent cells that are capable of forming cementum, lamellar bone, and fibrous tissue. Although central cemento-ossifying fibromas of the mandible are common, they are unusual in the maxilla. It is usually seen in middle age between third to fourth decades but rarely occurs at young age. Etiology is unknown but as per literature cemento ossifying fibroma in the bone might be caused by an irritant stimulus as reported. As numerous histological techniques have been used to identify the nature of calcification such as special stain and Immunohistochemistry, but there have been no cases reported using Gallego's stain which is a differentiating stain for hard tissue. This article reports a case of Juvenile cemento ossifying fibroma in 6 year old boy along with review of literature.

CASE REPORT

A 6 year old boy reported with a chief complaint of painless swelling over right side of the cheek since six months. The swelling was initially small in size which increased gradually without any discomfort to the present size. There was a history of road traffic accident at the age of one year at chin region and was operated for the same at the age of two. There was no history of pus discharge or epistaxis. No paraesthesia was present. Vision was normal.

Extra orally an obvious facial asymmetry was present over the right side of maxilla. The well demarcated swelling measured approximately 8cmX6cmX4cm in size. The extension of the swelling anteroposteriorly was from right ala of the nose to the vertical line corresponding to the outer canthus of the eye. The superoinferior extension was from the infraorbital region to the angle and inferior border of the mandible. Right ala of the nose was elevated. There was discoloration of the skin by a diffuse brown macular patch (Figure 1 a).

Intra orally all deciduous teeth were present except 53. A firm non-tender swelling was present extending antero-posteriorly from midline to maxillary tuberosity with bicortical expansion with obliteration of the buccal vestibule. Overlying mucosa showed areas of inflammation. Labial and buccal vestibule were obliterated (Figure 1 b).



Figure 1: (a): Extension of the swelling anteroposteriorly from right ALA of the nose to the vertical line corresponding to the outer canthus of the eye and superioinferiorly from the infraorbital region to the angle and inferior border of the mandible. right ala of the nose was elevated. (b): Swelling extending antero-posteriorly from midline to maxillary tuberosity with bicortical expansion with obliteration of the buccal vestibule. Overlying mucosa showed areas of inflammation. (c) & (d): 6 months follow-up, post operative healing was good.

Coronal section contrast enhance computed tomography passing through facial bones demonstrated, growth or mass in the right maxilla at the level of lateral aspect of nose. There was obliteration of nasal cavity of right side. (Figure 2 a) Axial section passing at the level of ramus of mandible showed a mass within the body of the maxilla. The lesion within the bone showed the density similar to that of a soft tissue and it is found that lesion is obliterating the right nasal cavity. Also, deviation of nasal septum marked asymmetry was noted onto the right side (Figure 2 b).

In the section taken at the level of maxillary sinus it was found that the lesion was obliterated and occupying maxillary sinus and deviation of nasal septum. Though the lesion was on the right side, there was a thickening of sinus lining on the left side. (Figure 2 c) Coronal section at the level of maxillary canine shows obliteration and effacement of right nasal wall. Density of the lesion was similar to that of the soft tissue. Floor of maxillary sinus and orbital floor was pushed superiorly (Figure 2 d).

Incisional biopsy was taken, hematoxylin and eosin stained section showed highly cellular connective stroma consisting of plump fibroblasts and collagen fibers.

Numerous concentric basophilic calcifications were seen along the ossifications having osteoblasts and osteocytes. Few blood vessels were seen and the features were suggestive of cemento-ossifying fibroma (Figure 3 a).

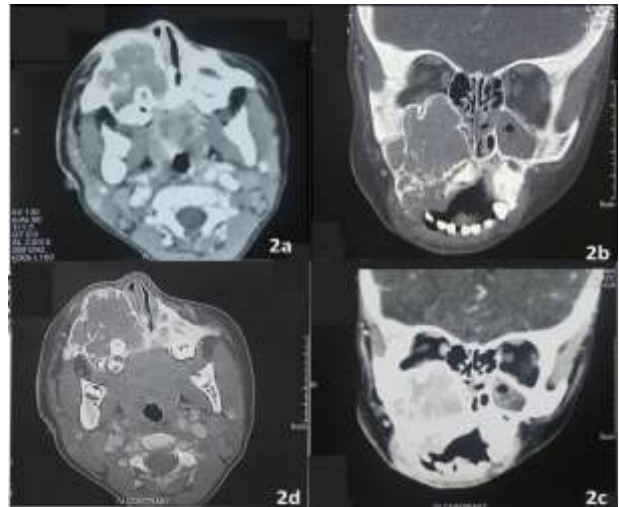


Figure 2: (a): Coronal section contrast enhance CT demonstrates mass in the right maxilla with obliteration of right nasal cavity. (b): Axial section passing at the level of ramus of mandible shows a mass with density similar to that of a soft tissue with deviation of nasal septum. (c): Section taken at the level of maxillary sinus shows lesion obliterating maxillary sinus with thickening of sinus lining on the left side. (d): Coronal section at the level of maxillary canine shows obliteration and effacement of right nasal wall. Floor of maxillary sinus and orbital floor was pushed superiorly.

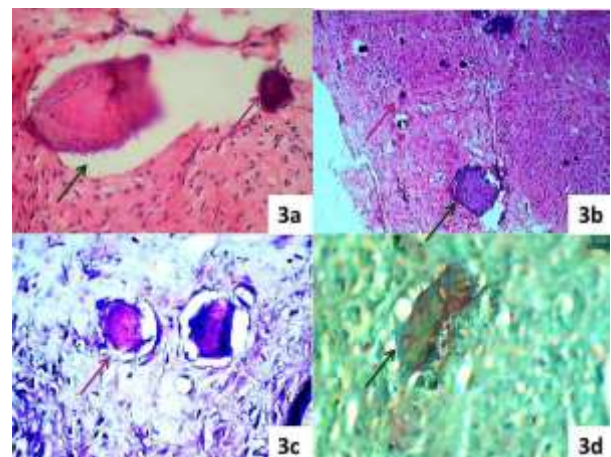


Figure 3: (a) & (b): Haematoxylin and eosin stained section showing cementum (↑) and bone (↑) depositions. (c) & (d): Gallego's staining showing entire cementum depositions (↑) stained dark pink in color and bony depositions (↑) stained green in color.

It was then excised and the tissue was sent for histopathological investigation. Hematoxylin and eosin stained tissue showed highly cellular connective tissue stroma with multiple areas of focal dystrophic calcifications. Connective tissue was highly fibrocellular with the plump fibroblasts. The dystrophic calcification was irregular, dystrophic, acellular and intensely basophilic. Multiple areas showed trabeculae of osteoid tissue (Figure 3 b).

Gallego's staining was also performed and it showed entire cementum depositions stained dark pink in color and bony depositions stained green in color (Figure 3 c, 3 d).

Diagnosis was given as juvenile cemento ossifying fibroma.

Patient was followed up after 6 months, post operative healing was good (Figure 1 c, 1 d).

DISCUSSION

In 1971 the World Health Organization (WHO) classified four types of cementum-containing lesions: fibrous dysplasia, ossifying fibroma, cementifying fibroma and cemento-ossifying fibroma. According to the second WHO classification, benign fibro-osseous lesions in the oral and maxillofacial regions were divided into two categories, osteogenic neoplasm and non-neoplastic bone lesions; cementifying ossifying fibroma belonged to the former category. However, the term "cementifying ossifying fibroma" was reduced to ossifying fibroma in the new WHO classification in 2005. Cemento-ossifying fibroma is a benign fibro-osseous-tumour.¹

The pathogenesis of COF is controversial. Earlier concepts relied on cementum, lamellar bone and fibrous tissue producing capacity of the pluripotent cells of periodontal ligament.² However lesions with similar histopathological features have been reported in other craniofacial structures and in long bones which do not contain cementum.³ Bone and cementum both are originated from mesenchymal stem cells. Accordingly, the classification of the World Health Organisation suggested the term ossifying fibroma and considered it as non-odontogenic neoplasm.⁴

COF is a disorder of unknown etiology. Bernier hypothesized that COF in the bone might be caused by an irritant stimulus (such as tooth extraction) which may activate the production of new tissue from the remaining periodontal membranes. The current theories regarding their origin include traumatic and developmental cases.⁵

Central cemento-ossifying fibromas occur more frequently in women than in men. They arise in the mandible in 62% to 89% of patients, 77% occurring in premolar region. Most are diagnosed between 20 and 40 years of age.⁶ When this tumor arises in children, it has been named as

the juvenile aggressive cemento-ossifying fibroma, which presents at an earlier age and is more aggressive clinically and more vascular at pathologic examination.⁷ The lesion in present case can be considered as rare and atypical based on its gender, age and site.

Apart from routine hematoxylin and eosin staining, Manish et al in 2012 performed special staining using Masson Trichrome stain for POF.⁸ In present case, Modified Gallego's Iron Fuschin stain was used which is a differential stain for cementum.⁹ Modified Gallego's stain is a variant of Lille's stain that uses basic reagents hematoxylin, carbol fuschin and aniline blue. The purpose of the stain is differential staining of the hard tissues seen in tooth and other pathological lesions. It has been used only in normal teeth using both decalcified and ground sections. This stain has also been tried by Sandhya et al in various odontogenic tumors as a preliminary report. Our finding with modified gallego's stain showed dark pink coloured cementum and green coloured bone which correlates with the finding of study reported by Sandhya et al.¹⁰

Due to the good delimitation of the tumor, surgical removal and curettage is the choice of treatment. In the case of very large lesions with important tissue ablation, the challenge is to replace the affected tissue. The prognosis is usually good, as recurrences are not frequent.¹¹

CONCLUSIONS

Therefore modified gallego's stain could be considered as better stain to differentiate hard tissue depositions in various pathologies.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: The study was approved by the Institutional Ethics Committee

REFERENCES

1. Y Liu, M You, H Wang, Z Yang, J Miao, K Shimizutani, et al. Ossifying fibromas of the jaw bone: 20 cases. *Dentomaxillofacial Radiology.* 2010;39:57-63.
2. Krausen AS, Pullon PA, Gulmen S. Cementomas--aggressive or innocuous neoplasms? *Arch Otolaryngol.* 1977;103:349-54.
3. Eversole R, Su L, ElMofty S. Benign fibro-osseous lesions of the craniofacial complex. A review. *Head Neck Pathol.* 2008;2:177-202.
4. Sirin Y, Kahraman N, Olgac V. Maxillary Cemento-ossifying Fibroma in a Male Patient with End-Stage Chronic Renal Disease. *Int. Journal of Clinical Dental Sciences.* 2011;2(3).
5. More C, Thakkar K, Asrani M. Cemento-ossifying fibroma. *Indian Journal of Dental Research.* 2011;22(2).

6. Reddy JK, Ramlal G, Dayanandam, Patil R. Cemento-ossifying fibroma of the Mandible: A Case Report And Review Of Literature. *Indian J Stomatol.* 2011;2(3):197-9.
7. Bertrand B, Ely P, Cornelius JP, Gosseys S, Clotuche J, Gillard C. Juvenile aggressive cemento-ossifying fibroma: case report and review of the literature. *Laryngoscope.* 1994;103:1385-90.
8. Sharma M, Sharma GK. Peripheral Ossifying Fibroma – A Masquerade. *Annals of Dental Research.* 2012;2(1):36-43.
9. Levey RJ. A Modified Gallego's Iron Fuschin Stain as a Differential Stain for Cementum. *J DENT RES.* 1956;35:491.
10. Tamgadge S, Tamgadge A, Srivastava C, Satheesan E, Bhalerao S. Modified Gallego's Stain as Differential Stain for Oral Hard Tissues in Oral Pathology: A Preliminary Report. *International Journal of Oral and Maxillofacial Pathology.* 2014;5(2):2-6.
11. Silvestre-Rangil J, Silvestre FJ, Requeni-Bernal J. Cemento-ossifying fibroma of the mandible: Presentation of a case and review of the literature. *J Clin Exp Dent.* 2011;3(1):e66-9.

Cite this article as: Tamgadge A, Tamgadge S, Venkateshwar G, Tiwari N, Dhauskar S. Juvenile cemento-ossifying fibroma: a case report using modified Gallego's staining. *Int J Res Med Sci* 2016;4:658-61.