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REVIEW ARTICLE

ANEURYSMAL BONE CYST IN CENTRAL OSSIFYING FIBROMA: "LESION IN A LESION", MANDIBLE

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ABSTRACT

Aneurysmal bone cysts (ABC's), a rare distinct pathologic entity, is an intraosseous, osteolytic lesion seen as locally destructive, rapidly expansile which has been reported to affect mainly the metaphyseal region of long bones and vertebrae. Aneurysmal bone cysts secondary to ossifying fibroma remains a relatively uncommon finding in the facial bones, and it is extremely rare with only very few cases reported in the mandible. This case report reveals one such, in a 42 year old lady, who presented with a solitary swelling of the right mandible, with obliterated vestibular depth, which showed Central Ossifying Fibroma as a pre-existing lesion, transforming into Aneurysmal Bone Cyst. Such rare entities are usually misdiagnosed and this gradually results in inappropriate treatment planning and poor prognosis.

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INTRODUCTION

Fibro-osseous lesions aim at replacement of normal bone architecture with benign fibrous tissues constituting fibroblast and collagen, various degree of mineralization. Several entities like ossifying fibroma, fibrous dysplasia and so on are included here. (Su *et al*., 1997) Various terms have been applied to these benign fibro-osseous neoplasms over the years. When bone predominates in a particular lesion, it is called an ossifying fibroma. Tumors containing both bone and cementum like material, with or without psammoma like bodies, a diagnosis of cemento-ossifying fibroma is made. (Kramer *et al*., 1992) Aneurysmal bone cysts are relatively rare, osteolytic lesion. Predominantly they are found within long bones (Rosenberg *et al*., 2005) and only 2% occur in the jaws (JABC's). (Motamedi and Yazdi, 1994) Jaffe & Lichtenstein recognised this cystic entity for the first time in 1942. (Jaffe and Lichtenstein, 1942) Since then, much debate and confusion has ensued regarding its nature and pathogenesis. An Aneurysmal Bone Cyst can occur as a secondary change in association with a number of benign and malignant lesions. (Munir J Nasser and Mohamed Shawarbi, 2010) Mandibular lesions are uncommon and can be mistaken for an odontogenic cyst. (Solomon *et al*., 2009) This report includes a transformation of Central Ossifying Fibroma into Aneurysmal Bone Cyst, an extremely rare entity that has been reported.

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Case report

A 42 year old lady complained of a gradually increasing swelling on the right side of her face since 10 months. Initially the swelling was small but gradually increased in size during past three months. Patient gives a history of trauma a year back in that region. Extraoral examination revealed the swelling extending anteroposteriorly from corner of the mouth to the angle of mandible, measuring around 4.5cms in size and superoinferiorly from ala-tragal line to 2cms below the lower border of mandible, measuring about 4.5cms in size. (fig 1) There was stretching of the skin overlying which was of normal colour as that of surrounding skin. Consistency was found to be firm and on palpation it was tender. There was no rise in temperature, no lymph nodes were palpable. Intraorally, a diffuse swelling was present extending superoinferiorly from marginal gingiva to the depth of vestibular sulcus measuring 3cms in size. It extends anteroposteriorly from distal aspect of 44 to mesial surface of 48, measuring 4.5cms in size. Interdental gingiva was found to be erythematous in relation to 45,46. On palpation, swelling was found to be tender and firm, with a bony hard consistency. Teeth were firm and vital. Obliteration of buccal sulcus with expansion of both buccal and lingual cortical plates was found. Orthopantamograph (OPG) (fig 2a) showed a solitary, unilocular radiolucency extending to the apical third involving both roots of 46 and inferiorly to the lower border of mandible. It approximately measured about 2cms x 3cms in dimension and was surrounded by well delineated and scalloped borders. Ct scan

shows bone loss with expansion of buccal and lingual cortex. (fig 2b) Provisionally, lesion was diagnosed as a traumatic bone cyst and clinicoradiograhic differential diagnosis included ameloblastoma, central giant cell granuloma or aneurysmal bone cyst. Haematological investigations revealed all values were under normal physiological limits. The lesion was provisionally planned for an incisional biopsy and the specimen was duly sent to the laboratory for a confirmatory diagnosis. Incisional biopsy reports were suggestive of Aneurysmal Bone Cyst, hence surgical intervention was planned accordingly.(fig 4a,b)

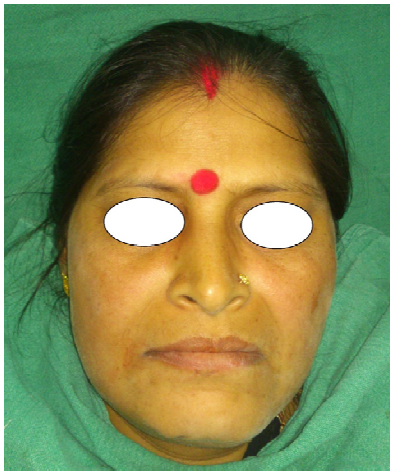


Figure 1. Pre-operative photograph, front profile

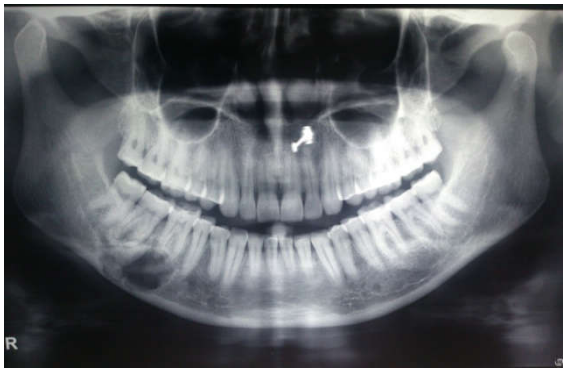
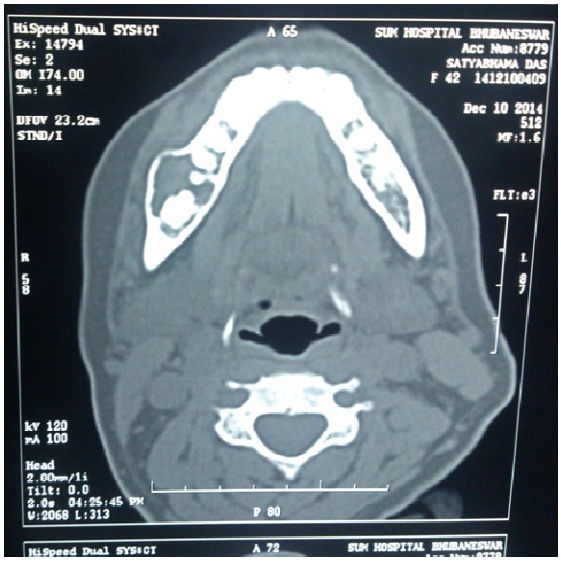


Figure 2: a) Pre-operative Orthopantomogram, showing mixed radiopaque and radiolucent lesion in 46 periapical region



b) Pre-operative CTscan suggestive of bone loss and expansion of buccal and lingual cortex

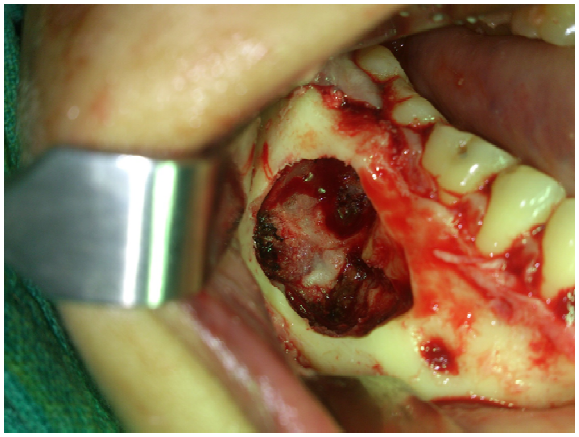
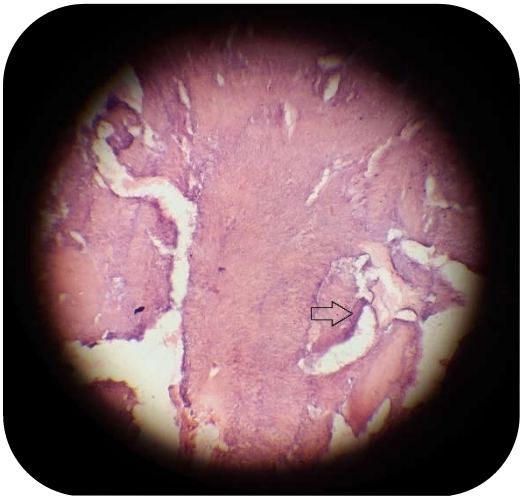
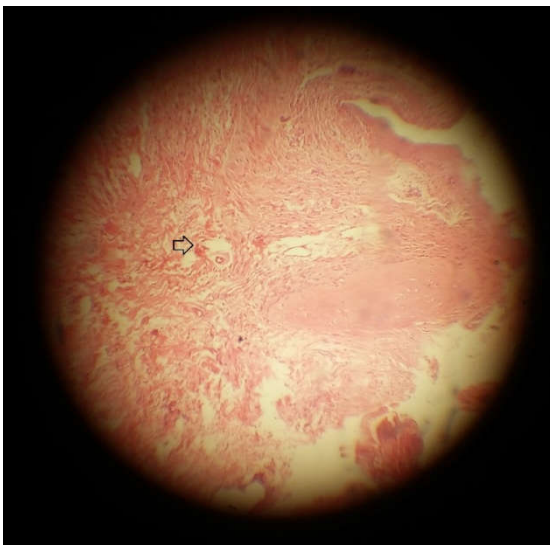


Figure 3. Intra-operative photograph showing multiloculed blood filled bony septae

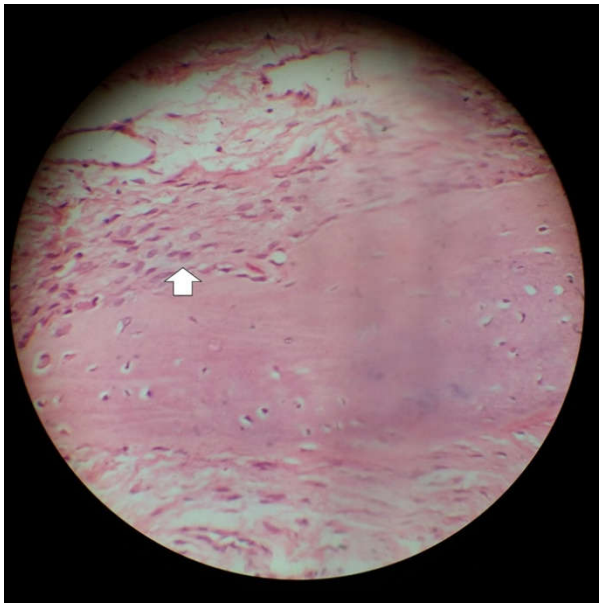


a)

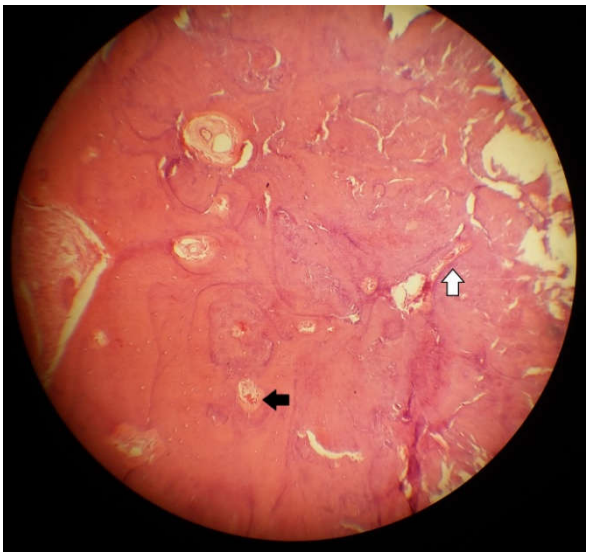


b)

Figure 4:
a) (H&E staining, original magnification x 100) Photomicrograph of the resected specimen of the mandible showing peripheral mature bony trabeculae (arrow showing)
b) (H & E staining, original magnification x 400) photomicrograph shows presence of giant cells with mild degree of inflammatory cells infiltration and presence of immature bone with varying size of spaces filled with unclotted blood.(arrow showing)



a)



b)

Figure 5:
a) (H&E staining, original magnification x 400) photomicrograph of the resected specimen shows collagenous stroma with numerous plump active fibroblast (white arrow)
b) (H & E staining, original magnification x 400) photomicrograph showing immature woven bone formation and mature bony trabeculae most of which are lined by osteoblasticrimming (white arrow), areas of large vascular channels are seen. Many extravasated RBC's are seen in stroma (black arrow)

During surgical procedure, on entrance the lesion excessive bleeding was encountered. The interior of the lesion showed a heterogeneously enhancing soft tissue mass with multiple bony septae.(Fig 3) A larger bone like mass was seen on the outer aspect, fragments of the cortical plate adherent to solid soft tissue. Multiple locular blood filled architecture was revealed. Multiple fragments were processed. Microscopically, H & E stained tissue sections showed collagenous stroma with numerous plump active fibroblast with many areas showing immature woven bone formation & mature bony trabeculae most of which are lined by osteoblastic rimming. Areas of large vascular channels are also seen. Many extravasated RBC's are seen in the stroma.(Fig 5a,b) The lesion was finally diagnosed as an Aneurysmal Bone Cyst which was secondary to Central Ossifying fibroma. Follow up was done after 3 months which showed bony healing at the lower border of mandible.

DISCUSSION

Benign fibro-osseous lesions of craniofacial complex are represented by variable disease processes characterized by pathologic ossifications and calcifications in collaboration with a hypercellular fibroblastic marrow element. (Roy Eversole *et al*., 2008) These lesions include fibrous dysplasia, ossifying fibroma and cement-osseous dysplasia. According to WHO, ossifying fibroma is classified into two types: the conventional/classic type & the aggressive type (juvenile ossifying fibroma). The later is further divided into aggressive trabecular and aggressive psammomatoid subtypes. (Foss and Fielding, 2007) These tumours are thought to arise from periodontal ligament and are composed of varying amounts of cementum, bone and fibrous tissue. Eversole *et al* reported association of the produced cementum like structures with membranous bone and may not only be related to cementogenesis. (Eversole *et al*., 1985) These lesions are slow growing and are most often seen in women between the third and fourth decades of life. (Tamiolakis *et al*., 2005) Asymptomatic instances being 50%, the growth of the tumour leads to facial asymmetry over time, mass causing discomfort/mandibular expansion and possible displacement of roots. (Sanchis *et al*., 2004) Although the underlying cause is not known yet, most of cases in literature have been found to have a history of trauma in the locus of the lesion. In accordance with the data found in the literature, our patient reported with a history of trauma in the affected area a year ago. Thus, it throws light on trauma as a possible triggering factor, postulating the lesion to be a connective tissue reaction rather than a neoplasm. (Feller *et al*., 2004) An Aneurysmal Bone Cyst is a non-odontogenic, non-epithelial cyst which commonly occurs in long bones and spine. (Mervyn Shear and Paul M Speight, 2007) The World Health Organization defined ABCs as expansive osteolytic lesions consisting of blood-filled spaces and channels divided by connective tissue septa that can contain osteoid tissue and osteoclast-like giant cells. According to Bruce *et al*, only 2-3% of the cases occur in head and neck region and among them, 66% of the cases occur in jaws. (Bruce H Matt, 1993) They commonly occur in the first two decades of life and do not show any gender predilection. (Mohammad HoseinKalantarMotamedi *et al*., 1993) Mandible is prone to be involved. More than 90% of the JABC's occur in the posterior regions of jaw. (Struthers and Shear, 1984) JABC's have different clinical pictures ranging from asymptomatic lesions occasionally discovered as radiolucencies on routine radiography to sometimes expansive and destructive patterns. (Kaffe *et al*., 1999) Painless swelling has been the main symptoms of JABC's according to literature reviews. (Biesecker *et al*., 1970) Patients with long bones ABC's frequently present with pain combined with a rapid growth pattern. (Szendroi *et al*., 1992) Patients can present with varying degrees of limited mouth opening, loosening of teeth, nasal obstruction and lip anaesthesia. (Park *et al*., 2008; Hardee *et al*.,1992) Aneurysmal Bone Cyst can develop as a secondary change in number of benign and malignant bone lesions. (Carolina AmaliaBarcellos Silva *et al*., 2011) On auscultation, no pulsatile sound will be detected over the tumour. Aspiration can result in fluid and is possibly an important diagnostic

method. The radiographic features of JABC's are variable. (KalantarMotamedi, 1998; Medeiros *et al*., 1993) Almost in many instances the lesion frequently appears as a well-circumscribed multilocular or unilocular radiolucency with a bloated appearance. Thin septa present might result in a soap bubble appearance. The teeth will be vital, with displacement and resorption of roots. (Lopez-ArcasCalleja *et al*., 2007) Histologically, other pre-existing entities decide the incidence of primary and secondary categories of ABC's. (Levy *et al*., 1975; Nadimi *et al*., 1987) Previously reported few cases reveal a very close association of Ossifying fibroma with JABC's as observed in this case report. (Arden *et al*., 1997; Citardi *et al*., 1996) The pathogenesis of ABC's remain controversial, with theories ranging from a post traumatic, reactive vascular malformation to genetically predisposed bone tumours. Most widely accepted theories to date has been that local circulatory abnormalities led to increased venous pressure resulting in dilation of local vascular network. Treatment concerns many methods like surgery, embolization, cryotherapy and a wait & see strategy. To date surgical therapy is still the most frequently applied treatment of ABC's secondary to Ossifying fibroma. The extent of surgery is related to the size and position of lesion, ranging from sample curettage to extended resection. (Cottalorda and Bourelle, 2007; Bernier and Bhaskar, 1958) Struthers and shears have proposed high recurrence rates of ABC's, usually recurring within the first year after the initial treatment.

Conclusion

Central Ossifying fibroma is a fibro-osseous lesion which occurs rarely in the mandible & when associated with Aneurysmal Bone Cyst. Early accurate diagnosis and appropriate treatment is necessary. Untreated cases result in extension of lesion into nasal, orbital and cranial cavities is common. Pretreatment biopsy and CT Scans are important tools for proper diagnosis and treatment plan. Due to high recurrence rate, long term follow up is necessary.

REFERENCES

Arden RL, Bahu SJ, Lucas DR. 1997. Mandibular aneurysmal bone cystassociated with fibrous dysplasia. *Otolaryngol Head Neck Surg*., 117:S153.

Bernier JL. and Bhaskar SN. 1958. Aneurysmal bone cysts of the mandible. *Oral Surg Oral Med Oral Pathol*., 11:1018.

Biesecker JL, Marcove RC, Huvos AG, *et al*. 1970. Aneurysmal bone cysts: A clinicopathologic study of 66 cases. Cancer 26:615.

Bruce H Matt. 1993. Aneurysmal bone cyst of the maxilla: case report and review of the literature. *International Journal of Pediatric Otorhinolaryngology*, 25: 211-26.

Carolina AmaliaBarcellos Silva, DDS, Alessandra Dutra Silva, DDS, José Antonio de CarvalhoSoares, DDS, Ney Soares de Araújo, DDS, PhD, Vera Cavalcanti de Araújo, DDS, CristianeFuruse, DDS, PhD, 2011. Trabecular Juvenile Ossifying Fibroma with Aneurysmal bone cyst: A Rare Presentation; Pediatric dentistry. Vol 33 / no 5 11.

Citardi MJ, Janjua T, Abrahams JJ, *et al*. 1996. Orbitoethmoid aneurysmalbone cyst. *Otolaryngol Head Neck Surg*., 114:466.

Cottalorda J. and Bourelle S. 2007. Modern concepts of primary aneurysmalbone cyst. *Arch Orthop Trauma Surg*., 127:105.

Eversole LR, Leider AS, Nelson K. 1985. Ossifying fibroma: A clinicopathologicstidy of sixtyfourcases. *OralSurg Oral Med Oral Pathol*., 60:505–11. (PubMed: 3864113)

Feller L, Buskin A, Raubenheimer EJ. 2004. Cementoossifyingfibroma:case report and review of the literature. *J IntAcadPeriodontol*., 6:131–5. (PubMed: 15553980)

Foss R and Fielding CG. 2007. Juvenile psammomatoid ossifying fibroma *Head and Neck Pathol*., 21 (1) 33–34 DOI: 10.1007/s12105-007-0001-x

Hardee PS, Whear NM, Morgan PR. 1992. Aneurysmal bone cyst ofthe maxilla—An association with tooth resorption. *J CraniomaxillofacSurg.,* 20:266.

Jaffe HL, Lichtenstein L. 1942. Solitary unicameral bone cyst with emphasis on the roentgen picture, the pathologic appearanceand pathogenesis. *Arch Surg*., 44:1004.

Kaffe I, Naor H, Calderon S, *et al*. 1999. Radiological and clinical features of aneurysmal bone cyst of the jaws. *DentomaxillofacRadiol*., 28:167.

KalantarMotamedi MH. 1998. Aneurysmal bone cysts of the jaws:Clinicopathological features, radiographic evaluation and treatment analysis of 17 cases. *J Craniomaxillofac Surg*., 26:56.

Kramer IRH, Pindborg JJ, Shear M. 1992. Histological Typing of Odontogenic tumours. 2d ed. Berlin:Springer-verlag.

Levy WM, Miller AS, Bonakdarpour A, *et al*. 1975. Aneurysmal bonecyst secondary to other osseous lesions: Report of 57 cases. *Am J ClinPathol*., 63:1.

Lopez-ArcasCalleja JM, CebrianCarretero JL, Gonzalez MJ, et al. 2007. Aneurysmal bone cyst of the mandible: Case presentationand review of the literature. *Med Oral Patol Oral Cir Bucal.,* 12:E401.

Medeiros PJ, Sampaio R, Almeida F, *et al*. 1993. Aneurysmal bone cyst ofthe maxilla: Report of a case. *J Oral MaxillofacSurg.,* 51:184.

Mervyn Shear, Paul M Speight. 2007. Cysts of the oral and maxillofacial regions. 4th ed.Blackwell Munksgaard Publishers; 150-55.

Mohammad Hosein Kalantar Motamedi, Finanavi, Pooyansadreshkevari, SeyedMehdijafari, Mohammad Ghasem Shams, Maryam Taheri, FatemehMashhadi Abbas, PouryaMotahhari. 1993. Variable presentations of Aneurysmal bone cyst of the jaws: 51 cases treated e H. Matt. Aneurysmal bone cyst of the maxilla: case report and review of the literature. *International Journal of Pediatric Otorhinolaryngology*, 25: 217-26.

Motamedi MH, Yazdi E. 1994. Aneurysmal bone cyst of the jaws:Analysis of 11 cases. *J Oral MaxillofacSurg*., 52:471.

Munir J Nasser and Mohamed Shawarbi, 2010. Hybrid psammomatoid ossifying fibroma & aneurysmal bone cyst of frontal sinus *J Pan Arab NeuroSoc*., 14(1) pp 97–9

Nadimi H, Bronny AT, Sbigoli A, *et al*. 1987. Aneurysmal bone cystassociated with a dentigerous cyst: Report of case. *J Am DentAssoc*., 115:859.

Park W, Nam W, Park HS, *et al*. 2008. Intraosseous lesion in mandibularcondyle mimicking temporomandibular disorders: Reportof 3 cases. *J Orofac Pain*, 22:65.

Rosenberg AE, Nielsen GP, Fletcher JA. 2005. Aneurysmal bone cyst,in Fletcher CDM, Unni KK, Mertens F (eds): WHO Classificationof Tumors: Pathology and Genetics of Tumors of SoftTissue and Bone (ed 3). Lyon IARC, p 338-339

Roy Eversole, Lan Su, Samir ElMofty, 2008. Benign Fibro-Osseous Lesions of the Craniofacial Complex A Review. *Head and Neck Pathol*., 2:177–202.

Sanchis JM, Peñarrocha M, Balaguer JM, Camacho F. 2004. Fibroma cementoosificante mandibular:Presentación de dos casos y revision de la literatura. *Med Oral*, 9:69–73. (PubMed: 14704620)

Solomon M *et al*. 2009. Psammomatoid juvenile ossifying fibroma of the mandible: A Histochemical insight *Internet J Dent Sci.,* 7 2

Struthers PJ, Shear M. 1984. Aneurysmal bone cyst of the jaws. (I).Clinicopathological features. *Int J Oral Surg*., 13:85.

Su L, Weathers DR and Waldron CA. 1997. Distinguishing features of focal Cemento-osseous dysplasia and cemento-ossifying fibromas: I. A Pathologic spectrum of 316 cases *Oral surg Oral Med Oral Pathol Oral RadiolEndod*., 84(3) 301–9 DOI: 10.1016/S1079-2104(97)90348-6 PMID: 9377196

Szendroi M, Cser I, Konya A, *et al*. 1992. Aneurysmal bone cyst: Areview of 52 primary and 16 secondary cases. *Arch OrthopTrauma Surg.,* 111:318.

Tamiolakis D, Thomaidis V, Tsamis I. 2005. Cementoossifying Fibroma of theMaxilla: a Case Report, *ActaStomatol Croat*, 39:319–21.

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