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CASE REPORT

Aneurysmal Bone Cyst: Exploring the Diagnostic Dilemma of Un-yielding Jaw Cyst and Its Management

Sakshi Batra[1] , Adit Srivastava[2], Chandresh Jaiswara[3], Arjun Mahajan[4], Harsha Popli[5],

[1]Post Graduate student, Oral Medicine and Radiology Unit, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, India, ORCID ID: 0000-0001-7496-3289

[2]Professor, Oral Medicine and Radiology Unit, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, India, ORCID ID: 0000-0001-7660-4650

[3]Professor, Oral and Maxillofacial Surgery Unit, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, India, ORCID ID: 0000-0002-7499-1359,

[4]Post Graduate student, Oral and Maxillofacial Surgery Unit, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, India, ORCID ID: 0000-0001-9475-9357

[5]Senior Resident, Oral Medicine and Radiology Unit, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi, India, ORCID ID: 0000-0001-7739-4528

Abstract:

An aneurysmal bone cyst (ABC) is a rare benign, blood-filled bone lesion that most commonly occurs in the spine and metaphysis of long bones, most notably the femur, and tibia. Metaphysis of long bones account for 12% and 30% of the spine. However, 2% and 12% of ABCs occur in maxillofacial skeleton. In the maxillofacial region mandible is most commonly affected with a higher predilection for the molar area with average age of presentation being 13 years, with 80% cases being less than 20 years,however, there is no sex predilection. Because aneurysmal bone cysts of the head and neck region can manifest as a fast-growing, expanding, and destructive lesion, clinicians must be aware of this entity in order to appropriately identify and treat patients.We report here a case of ABC of the right mandibular region at early age of 9 years associated with history of trauma and follow-up of 1-year post surgery.

Keywords: Aneurysmal bone cyst, pseudocyst, radiolucent lesion.

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Corresponding author: Dr. Sakshi Batra, Junior Resident, Oral Medicine and Radiology Unit, Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University

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Introduction:

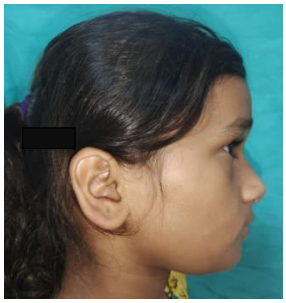
A benign pseudo-cystic osseous lesion characterized by a fibrous connective tissue stroma with cellular fibrous tissue, multinucleated giant cells, and huge blood-filled voids with no endothelial lining is known as an aneurysmal bone cyst (ABC).[1] First identified by Jaffe and Lichtenstein in 1942, is a non-neoplastic bone lesion that has been observed to mostly affect the skeleton's long bones.ABC in the long bones is distinguished radiologically by a well-defined expansile radiolucent lesion surrounded by a thin overlaying cortex. In contrast, reports of ABC in the jaws are conflicting, ranging from a mostly unilocular radiolucency to a 'ballooned out' multilocular radiolucency with a honeycomb or soap-bubble look.[2] ABC can arise as a main lesion or as a sequel lesion in 29-57% of cases, and it can be associated with a variety of different non-malignant or malignant precursor lesions. These precursor lesions are most commonly giant cell tumors (19-39%), but they can also be bony disorders such as osteogenesis imperfecta, fibrous dysplasia, chondroblastoma, chondromyxoid fibroma, non-ossifying fibroma, chondrosarcoma, fibro myxoma, fibrosarcoma, fibrous histiocytoma, radiation osteitis,

eosinophilic granuloma, osteoblastoma and osteosarcoma.[3,4]

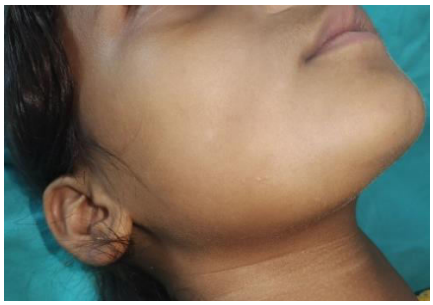
Its exact etiology and pathophysiology remain unknown. Although the ABC is not a real neoplasm, it does entail both a hemorrhagic and a hyperplastic process. Many theories on the origin of the ABC have been proposed throughout the years, including hemodynamic disturbance of the bone, trauma and subsequent hemorrhage, skeletal hemangioma, and arterio-venous fistula.[5] ABCs are often asymptomatic, however they might present with local discomfort and swelling. Only 2-3% of ABCs occur in the head and neck region, with the mandible and maxilla being the most prevalent occurrence sites. Aneurysmal bone cysts are more common in women and appear in the first few decades of life.[8]Correlation of clinical, radiological features are necessary but final diagnosis is based on histopathological reports.This case represents a mandibular aneurysmal cyst showing ballooning of cortex in radiographs in a young patient of 9 years, along with basic etiology known till now of trauma.The patient was followed up for 1 year and showed healing of the lesion with almost complete bone formation radiographically.

Case Report:

A 9-year female patientvisited the Department of Oral Medicine and Radiology Department with the chief complaint of swelling on right side of face since 3-4 months which was not appreciated initially but increased gradually to the present size. [Fig.1,2]

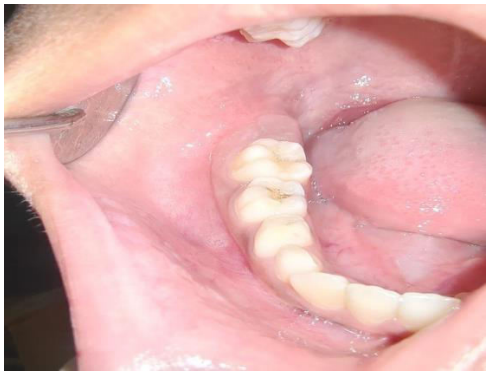


[Fig.1.Lateral view of the patient with mild swelling at initial visit.]



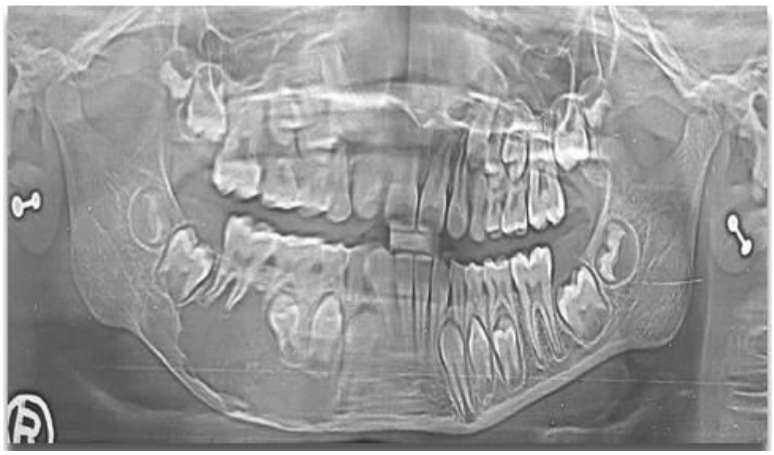
[Fig.2.Right mandible swelling]

There was no history of pain, bleeding, pus discharge or fever. No history of any deleterious habit or any difficulty in speaking and chewing. However, history of trauma due to fall on ground 1 year back was reported by the patient's attendant. On extra-oral examination mild well-defined swelling was present on the right side of the jaw extending from the lower border of the mandible to mid of the body of the mandible superio-inferiorly and antero-posteriorly in mid of body of the mandible approximately 3cmx2.5cm, hard in consistency, with smooth surface,non-tender, and non-compressible.On Intra-oral examination no appreciable swelling was seen, however mild tenderness was present on the buccal vestibule with respect to 85,46 and 46 was vital.hence, based on the above findings provisionalof dentigerous cyst was made.[Fig.3]Further investigations were carried out and on aspiration blood-tinged fluid which was non-diagnostic [Fig.4]. Orthopantomogram showed a multi-locular radiolucency with radiopaque flecks in between, along with irregular borders extending with respect to 46 and developing 44,45,47.[Fig.5]



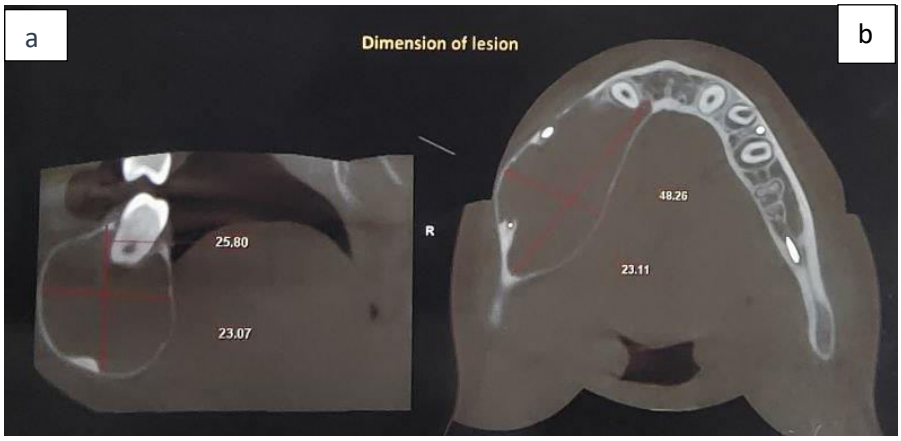
[Fig.3.Intra-oral view of the patient at initial visit, with no swelling and carious deciduous mandibular second molar]

[Fig.4.Aspirated fluid].



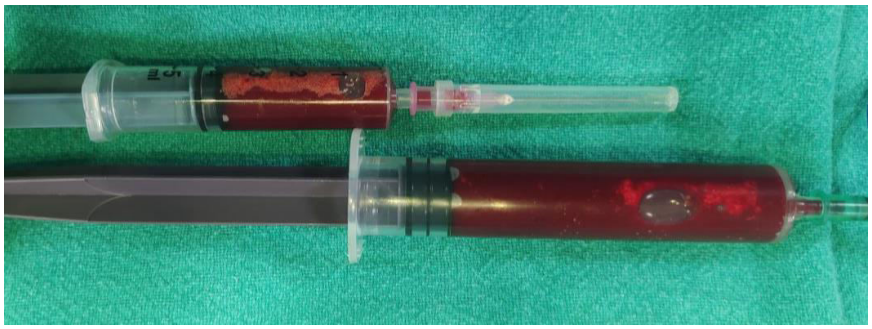
[Fig.5 Initial OPG of the patient]

Later on, CBCT (Cone beam computed tomography) showed a large radiolucency on right side of body of mandible measuring antero-posteriorly approximately 48mm,supero-inferiorly 25mm and medio-laterally 23mm along with buccal and lingual cortical expansion and thinning and IANC appeared to be displaced buccally.[Fig.6 (a) (b)]

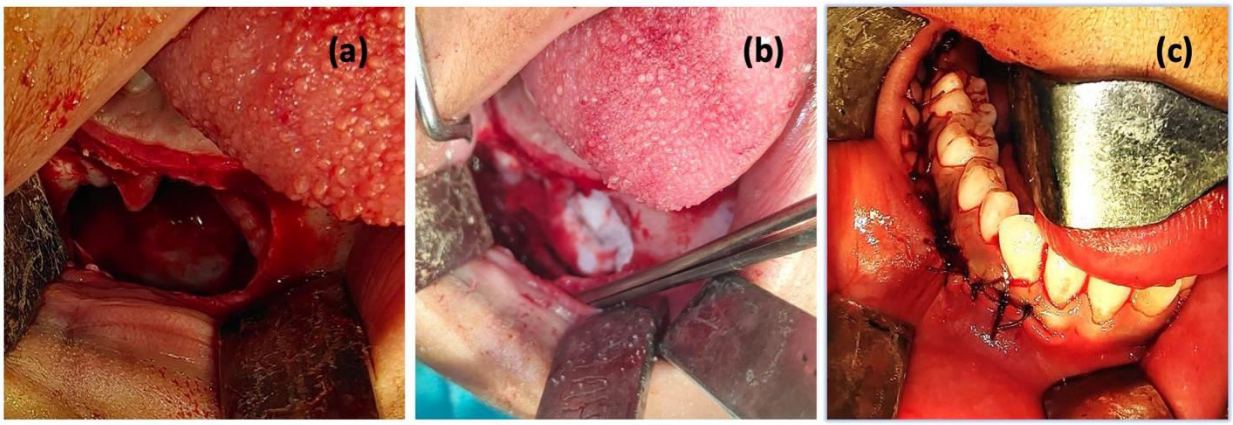


[Fig.6CBCT view showing supero-inferior dimension(a) and axial view showing antero-posterior and mediolateral dimension(b).]

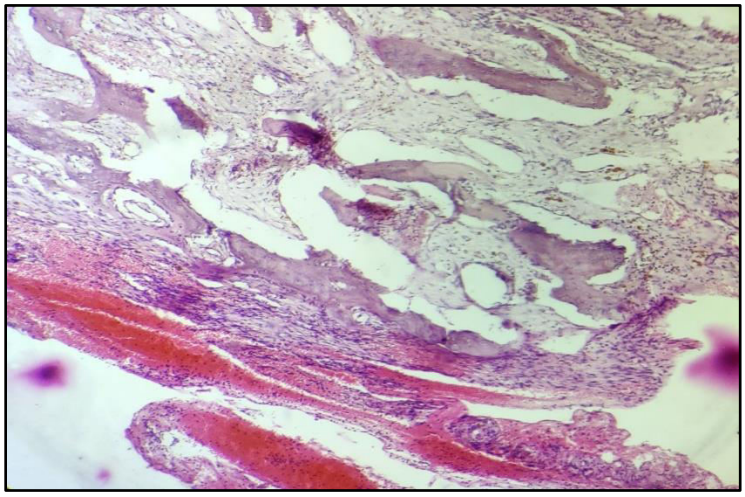
Based on these finding a radiographical differential diagnosis of central giant cell granuloma, aneurysmal bone cyst, ameloblastoma, Odontogenic keratocyst was made. With normal CBC, RBS, Chest X-ray and negative viral markers patient was planned for curettage or enucleation under general anesthesia. Taking care of all the aseptic precautions patient was painted with 10% betadine and draped with sterile linen. Throat packing was done, and surgical site was infiltrated with lignocaine and adrenaline (1:2,00,000). Crevicular incision was given and muco-periosteal flap was raised to expose the cavity filled with blood-tinged fluid[Fig.7] frombuccal aspect[Fig.8(a)]. Intralesional curettage followed by peripheral ostectomy was done with high-speed bur to disrupt the architecture of the lesion till fresh bleeding was seen. Irrigation was done with 10% betadine and saline and after placement of gel-foam [Fig.8(b)]closure was done with 3-0 vicryl[Fig.8(c)]. Patient was extubated with all vital within normal limits. Specimen of bone taken during peripheral ostectomy was send for histopathological examination.Low power view showed fibro-cellular connective tissue stroma with dense infiltrate of chronic inflammatory cells.CT stroma consists of bony trabeculae containing osteocyticlacunae, filled with osteocytes and outer osteoblastic rimming.Numerous endothelial-lined blood vessels with focal areas of hemorrhage and extravasated RBCs are seen.[Fig.9]



[Fig.7Contents of cystic cavity]

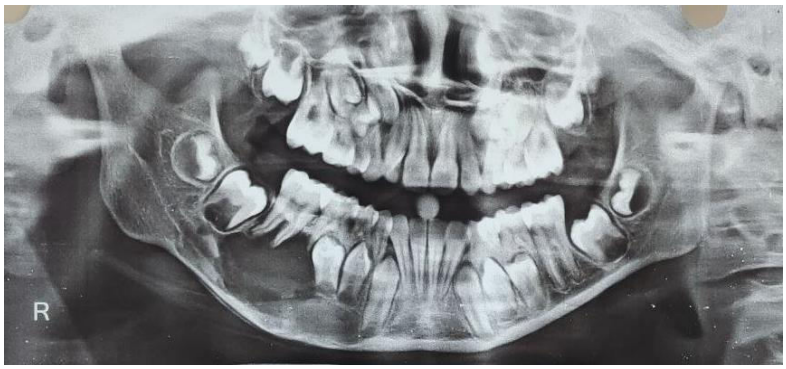


[Fig.8 (a) lesion after exposure, (b) gel-foam placement after peripheral ostectomy, (c) closure]

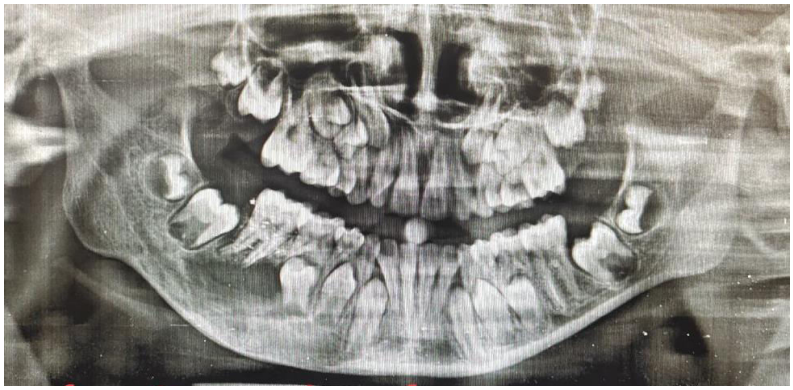


[Fig.9. Histopathological Low power view]

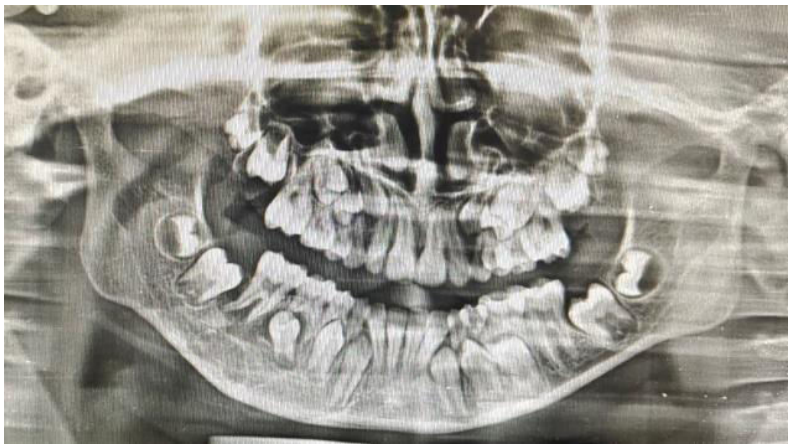
Relating to radiographical , clinical, surgical, and histological features a final diagnosis of Jaw Aneurysmal Bone Cyst was made,However as it is the most commonly occurring in long bones of body , for which various radiographs were taken and they were non-significant. Further followed up at1stmonth,6th month and 1year showing radio-opacity that represented the bone formation and hence healing of the cyst.[fig10,11,12]



[Fig.10 OPG AT 1ST month showing woven bone formation around 43,44]



[Fig.11OPG at 6th month with increased bone formation in the cavity.]



[Fig.12 OPG at 1year follow-up showing almost complete radiopacity in cystic lesion.]

Discussion:

A unicameral cyst is an aneurysmal bone cyst (ABC). Van Arsdale appears to have described this item as an ossifying hematoma in 1893. Bloodgood described a condition called a periosteal hematoma in 1910. Barrie coined the term hemorrhagic osteomyelitis in 1922. [5]Other authors have observed the resemblance to giant cell tumours and have used terminology such giant cell tumour of bone, atypical giant cell tumour, and subperiosteal giant-cell tumour to describe it. Other names for this condition include osteitis fibrosa cystica, expansile hemangioma, and aneurysmal giant cell tumour. [1,2,3]

Various ideas have been proposed to explain the pathophysiology of ABCs from the initial report of JABC, the earliest being that a localized circulatory anomaly caused increased vascular pressure and vasodilatation of the local vascular network, resulting in ABCs.[4]Another notion portraying ABCs as a reactive process gained traction as a result of several reports of ABCs occurring concurrently with or within neoplasms, both benign and malignant. Some investigations even

stated that ABCs should only be treated as a secondary lesion, with the underlying or pre-existing lesion being overlooked or overlapped by morphologic changes into ABC.[5,9]

Recently, chromosomal changes in regions 17p and 116q were discovered, implying a neoplastic etiology for the lesion. Approximately 74% of the instances with chromosomal anomalies reported in the literature to date have involved chromosomes in some way.[8] Panoutsakopoulos et al. described 3 cases of ABCs with chromosomal anomalies, band 16q22 being involved in all 3 patients. Other authors have subsequently demonstrated several different rearrangements involving different chromosomes such as 1, 2, 6, and 11.[6,7,14]

The mandible is the most commonly affected with a higher predilection for the molar areas. The average age of presentation is 13 years, 80% of patients are less than 20 years old and there is no sex predilection. ABC's clinical presentation is not specific. It might range from a minor, localized lesion to an aggressive, rapidly destructive lesion perforating the bone's cortical plate. The surrounding oral mucosa is normal, and the lesion appears bluish under the microscope. Tooth dislocation and malocclusion are prevalent.[2,8]

Capanna et al classified the ABCs into three stages based on clinical and radiological findings. Inactive cysts have complete periosteal shells with distinct sclerotic margins. Cysts in the active stage have incomplete periosteal shells but distinct edges. Aggressive cysts have irregular borders and homogenous osteolysis. Whereas active or aggressive ABCs tended to reoccur in their series, inactive ABCs did not. This type of staging procedure is critical for treatment, yet it has never been acknowledged in JABC reports.[9,12]

ABCs can be seen on a radiograph. CT and MRI scans are critical for both diagnostic and surgical management of ABCs.Aneurysmal bone cysts show distinctive patterns on CT and MRI that help clinicians reach a diagnosis. ABCs show expansile, lucent bone disintegration with osseous remodeling and cortical thinning on CT. Fluid levels within the ABCs can be seen more clearly on MRI than on CT. MRI can also reveal a "soap-bubble" pattern caused by the cystic structure of ABCs, as well as outline internal septations of the ABC. While CT and MRI are useful in aiding in the diagnosis of ABC, a conclusive diagnosis can only be made histologically. ABCs are extremely vascularized under the microscope, displaying delicate blood-filled structures capillaries and distended venous channels. In between the vascular spaces, it is common to visualize islands of bone, fibrous tissue, and multinucleated giant cells.[2,10,11]

Curettage, block resection with reconstruction, therapeutic embolization, and open packing have all been utilized as therapies. All ABC procedures entail the total excision of the benign lesion. Removing a lesion that is restricted within bone is normally simple, but it can be challenging if the lesion is multilocular, expansile, separated by many bony septa, destructive, or the cortices are punctured.[7]

Curettage with or without bone graft is the standard of therapy for ABCs, depending on the size of the void. Despite the best curettage efforts, clinical series have revealed widely varied recurrence rates, with some series reporting rates as high as 59%. As a result, several adjuvants, such as cement, high-speed burr, argon beam, phenol, and cryotherapy, have evolved to reduce recurrence.In the hopes of reaching equal results with fewer consequences, some groups have examined less aggressive surgical procedures and medical managements such ascuroscopy, percutaneous doxycycline, Bisphosphonate medical therapy, RANKL inhibition, and the function of denosumab, etc.[3,13,15]

Conclusion:

ABCs are well-defined, expansile lesions that can thin and expand the cortical bone and are visible on radiographs with unilocular or multilocular appearance. The histopathologic analysis of a biopsy specimen yields a conclusive diagnosis.Surgical excision is the preferred treatment for ABCs, accomplished through curettage or excision of the lesion. Adjuvant therapy, such as radiation therapy or bone grafting, may be required in some circumstances. The prognosis for ABCs is excellent with complete surgical excision and the recurrence rate is low

This article's case study serves as an example of the clinical, radiological, and histopathologic characteristics of an ABC of the mandible. The patient's lesion was surgically removed with success, and there had been no recurrence at her one-year follow-up.In the differential diagnosis of a mandibular enlargement that causes no pain, this case report emphasizes the significance of taking ABC into account. ABCs must be diagnosed and treated as soon as possible in order to avoid problems including facial deformity and bone resorption.

Conflict of interest: Authors declares no conflict of interest.

Author contributions

All authors contributed to manuscript and design. Material preparation and were performed by Sakshi Batra[1], Adit Srivastava[2],Chandresh Jaiswara[3], Arjun Mahajan[4], Harsha Popli[5]

The first draft of manuscript was written by Dr. Sakshi Batra[1*]and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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