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Case Report

Dentistry Section

Solitary Bone Cyst of Maxilla in a 12-year-old Child: A Case Report

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ABSTRACT

Solitary Bone Cyst (SBC) are the bony cysts which lack epithelial lining. They are also known as simple bone cysts or pseudocyst. This fluid filled lesions, lies in an intact bony wall. These lesions are not a common clinical finding and are frequently discovered by chance in radiographs during routine examinations. The aetiopathogenesis has not been studied in depth, and the management remains controversial. When presented in oral cavity, the most common site of occurrence is body of mandible with 75% occurrence among jaw bones while prevalence in maxilla is only 1% in the jaw bone. In this case report, solitary bone cyst is presented in a 12-year-old male patient, who came with chief complaint of bony enlargement on buccal aspect of maxilla in the posterior region. The lesion presented as a painless swelling for three months gradually increasing in size. The patient was treated with surgical excision and there was no sign of regional recurrence at three months of postoperative follow-up. In this paper, the authors presented a rare case report of solitary bone cyst in maxilla in a paediatric patient.

Keywords: Bony lesions, Cysts in children, Cysts of oral cavity, Oral lesions, Simple bone cyst

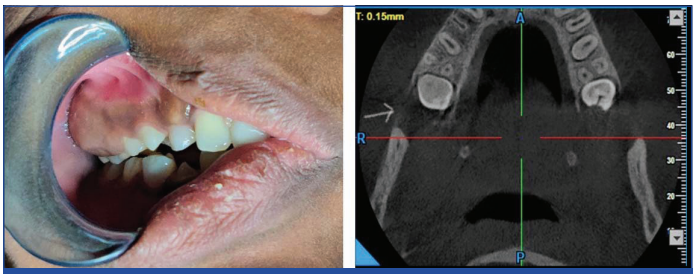
CASE REPORT

A 12-year-old male patient, reported to the Outpatient Department with the chief complain of an enlargement present on right side of the upper jaw since three months which was not associated with any pain but patient was concerned due to increasing size of the lesion as it was creating problems in masticatory functions and aesthetics [Table/Fig-1]. On taking history, it was revealed that patient did not have any history of trauma, also there were no associated symptoms present. Patient noticed that the swelling enlarged rapidly within two to three months from a small nodular growth to a large bony mass. There was no significant past medical history and no family history of similar occurrence. Patient was completely well on physical examination.

The extraoral examination of child revealed leptoproscopic facial form with mesocephalic head form. Lips were competent and no facial asymmetry was observed. The patient was otherwise healthy. Intraoral examination of oral cavity presented late mixed dentition and revealed a unilateral enlargement present on posterior aspect of maxillary bone on buccal cortical plate involving tooth number 14, 55, 16 and region posterior to tooth number 16 extending upto the maxillary tuberosity. The enlargement was bony hard on palpation, non tender, non mobile. The overlying mucosa was thin and blanched. The lesion had clear boundaries with a diameter of 1.5 cm. The teeth 11,12,13,14,16 were vital on conducting vitality tests using electric pulp tester.

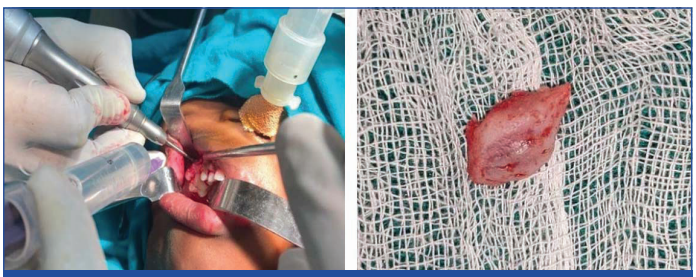
Also, there was no history of pain or sensitivity associated with any of the teeth adjacent to the area of lesion. Patient presented with Cone Beam Computed Tomography (CBCT), obtained from previous hospital, which showed a radiolucent unilateral lesion on the right side, confined to the posterior region of maxilla on the buccal aspect covering the roots of premolar and molar teeth, with defined borders [Table/Fig-2]. On the basis of radiological findings, the differential diagnosis was made as ameloblastoma, keratocystic odontogenic tumor, myxoma, and central giant cell granuloma. With available clinical and radiographic findings, the clinical diagnosis of SBC was made. After taking an informed consent, the patient was scheduled for excisional biopsy under general anaesthesia, the treatment was planned to remove the bony mass.

A full thickness flap was raised to expose the lesion adequately. Using a bone cutting carbide bur no. 702 SS white, the bony growth



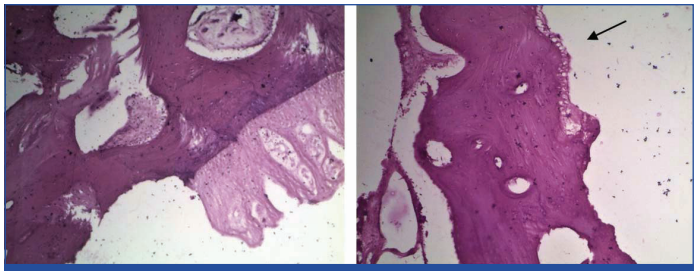
[Table/Fig-1]: Clinical presentation of lesion; blanching and thinning of mucosa can be observed. **[Table/Fig-2]:** CBCT showing radiolucent unilateral lesion on right side in posterior maxillary region. (Images from left to right)

was cut under continuous saline irrigation, using bone filer the rough edges of the surrounding bone were smoothened and the granulation tissues were curated using a curate. Once the smoothening is done, the area was washed thoroughly with iodine and saline solution in 1:1 proportion. The flap was closed with sutures which was followed by the placement of the periodontal dressing [Table/Fig-3]. Size of excised lesion was 1.5 cm in diameter [Table/Fig-4], the lesion was then sent for biopsy for the confirmation of pathology and final diagnosis. Histopathological examination showed the fragments of bony trabeculae with fibrous septa lacking any lining epithelium, these findings were consistent with the diagnostic histological features of SBC thus confirming the final diagnosis of solitary bone cyst [Table/Fig-5,6].



[Table/Fig-3]: Excisional biopsy performed under continuous saline irrigation. **[Table/Fig-4]:** Excised bony mass, 1.5 cm in diameter. (Images from left to right)

Antibiotics, analgesics and mouthwash were prescribed for next seven days and patient was kept on a liquid diet. No postoperative complications were noticed and patient was discharged the next day. Follow-up appointment was scheduled after 10 days of surgery



[Table/Fig-5]: Histopathological image with 10x magnification, using H&E stain. Bony trabeculae with fibrous septa without epithelial lining can be seen.
[Table/Fig-6]: Histopathological image with 100x magnification, using H&E stain. Bony trabeculae with fibrous septa without epithelial lining. (Images from left to right)

and suture removal was done. Tissue appeared healing and patient was asymptomatic after 10 days. The lesion healed uneventfully and no postoperative complications were seen after three months follow-up [Table/Fig-7,8].

[Table/Fig-7]: Follow-up after 10 days, healing tissues can be observed.
[Table/Fig-8]: Follow-up after three months, complete healing can be observed. (Images from left to right)

DISCUSSION

Solitary Bone Cyst (SBC) was described as an entity in 1929 by Lucas. The diagnostic criteria were first established in 1946 by Rushton. [1,2] This is an idiopathic cyst, also considered as a pseudocyst. Some other names include traumatic bone cyst, simple bone cyst, haemorrhagic bone cyst, haemorrhagic cyst, idiopathic bone cavity and unicameral bone cyst. The term "solitary bone cysts" was recommended by international [3]. SBC are intraosseous pseudocyst having a connective tissue lining without any epithelial lining, from inside it is either empty or filled with blood, serum, or serosanguineous liquid [2,4].

The SBC are commonly found in other parts of skeleton, normally in the medulla of long bones (90-95%) with a high prevalence in the proximal metaphyseal region of the humerus (65%) and the diaphyseal axis of the femur (25%) [5,6]. Only 10% occurs in the jaws; 75% of these occurs in the body of mandible, more frequently involving the posterior region and less common in the anterior mandible/mandibular symphysis. In the body of mandible, the second premolar is the most common location [5]. Only 1% of maxillary cysts constitutes solitary bone cysts, with most reported cases in the anterior region than posterior region and therefore, the presented case added an important contribution. The occurrence of SBC majorly happens during second decade of life without any sex discrimination [7,8,9]. However, some authors reported male predilection with male:female ratio of 1.6:1. Seen more commonly in Asian and black females according to Howey's analysis [10].

The lesions of SBC are commonly asymptomatic; however, some authors have reported complains of pain, tooth sensitivity and paresthesia associated with displacement of the inferior dental canal [10]. As reported by Forssell K et al, in 1988, 30% of SBC cases there was presence of pain in the affected area [11].

Three main theories have been speculated to explain the aetiopathogenesis of SBC as: a) abnormal osseous growth b) degenerating tumoral process c) A particular factor triggering haemorrhagic trauma [12]. However, the most accepted theory is intermedullary haemorrhage. In SBC the haemotoma fails to organize in marrow space leading to liquefaction of clot and necrosis of bone marrow which causes trabecular resorption by osteoclastic activity. Following this, there is increase in osmotic pressure which is due to the breakdown products of haemolysis, the increased osmotic pressure leads to transudation and cyst formation [13,14]. Therefore, most of the SBC develops after trauma or previous injuries. The spongy bone contains haemopoietic marrow and gets enclosed in heavy compact cortical layer, this explains the common occurrence of SBC in mandible and in young people and children [13].

Most widely accepted treatment for SBC is surgical exploration followed by curettage of the bony walls. The surgical exploration produces bleeding in the cavity and serves as diagnostic maneuver and a therapeutic therapy [15,16]. Resolution of the lesion depends upon size of the lesion and usually takes place in six months with a good prognosis and rare reoccurrence rate. Other approaches for the treatment include use of allogenic materials like lyophilized bone, hydroxyl-apatite or gel foam to fill the bone cavity in cases where conventional management fails and dental implant rehabilitation is required [17,18]. These cystic lesions are often asymptomatic without any complain of pain and diagnosed in routine radiographic examinations. Radiographically SBC presents radiolucent lesions mostly having well-defined margins and normal appearance [8].

It is very uncommon to see the presence of fistulas, root resorption, paresthesia, and pathological fractures in association with these lesions [18]. CBCT aids in authentic radiographic diagnosis of SBC if orthopantomography fails to do so. Major complications have not been reported with these cystic lesions but extensive lesions can cause pathological bone fractures [19]. The main radiographic feature is scalloping which is seen in the lesions that are extended towards the apex of roots. In extensive lesions, the cortical bone tends to be thinned due to intraosseous erosion without causing bone expansion or fracture [20].

In this case, the patient came with the chief complaint of unusual swelling on right side of the maxilla which gradually increased in size within two to three months. On clinical presentation the lesion was bony hard on palpation without any pain or tooth sensitivity. Also, patient did not provide any history of previous trauma. Surgical excision or curettage was the treatment of choice, therefore excisional biopsy of the lesion followed by curettage was chosen as treatment plan. Treatment was uneventful and the child did not present any complications after 3 months of follow-up. Similar case reports have been tabulated in [Table/Fig-9] [21-23].

S. No.	Author's name and year	Place of study	Age of subjects	Site of occurrence	Clinical findings	Diagnosis and treatment
1.	Patil SP et al., 2019 [21]	Department of Oral Medicine and Radiology, Rural Dental College, Ahmednagar	15-year-old female	Posterior maxilla	No clinically detectable lesion. On radiographic examination, panoramic view showed a 2 cm×1.5 cm well-defined unilocular radiolucent lesion with sclerotic border extending from the apical region of the upper right second premolar to the second molar. CBCT of maxilla (5 mA, 90 kV, and 360°) also revealed single, large, well-defined, expansile radiolucent lesion of size 2.8 cm×2.1 cm on the right side of the maxilla.	Solitary or traumatic bone cyst. Treatment: Patient was advised for conservative surgical excision, but patient did not show up.

2.	Saldaña SA et al., 2018 [22]	Mexico	45-year-old male	Right side of the upper jaw and both sides of the lower jaw.	A slow-growing, asymptomatic, Indurated lesion for 2-years. Clinically there was an increase in the right region of the upper jaw, measuring approximately 1.5 cm. Palpation of lower jaw revealed bilateral volume increase in the region of the mandibular body and chin area. Three dimenstional reconstruction of the facial mass revealed expansion and perforation of bilateral mandibular vestibular cortical plate, with no data of tooth displacement.	Solitary bone cyst on upper and lower jaw. Treatment: Surgical incision of cystic cavity followed by curettage of the affected area.
3.	Llobet L et al., 2019 [23]	University of Barcelona, Spain	15-year-old patient	Posterior region of mandible.	No clinical variation was present, lesion was detected in routine prosthodontic visit. The CBCT showed the presence of well-defined unilocular radiolucent lesion.	Solitary bone cyst of mandible. Treatment: Continuous surgical decompression was done
4.	Present study	Department of Paedodontics and Preventive Dentistry, Bilaspur, Chhattisgarh	12-year-old-male	Posterior region of maxilla.	Unilateral enlargement presents on posterior aspect of maxillary bone on buccal cortical plate involving tooth number 14, 55, 16 and region posterior to tooth number 16 extending upto the maxillary tuberosity.	Excisional biopsy was done under general anaesthesia and the sample was sent for histological examination.

[Table/Fig-9]: Similar case reports of solitary bone cyst [21-23].

CONCLUSION(S)

Solitary bone cyst is considered as a pseudocyst without any epithelial lining. In this article, authors reported a rare case of 12-year-old male child with SBC in posterior region of maxilla including clinical, histopathological and radiographic findings along with treatment modalities.

REFERENCES

[1] Lucas C, Blum T. Do all cysts of the jaws originate from the dental system. J Am Dent Assoc. 1929;16:659-61.
[2] Rushton MA. Solitary bone cysts in the mandible. Br Dent J. 1946;81:37.
[3] Wright JM, Vered M. Update from the 4th edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and maxillofacial bone tumors. Head and Neck Pathology. 2017;11(1):68-77.
[4] Barnes L, Eveson JW, Reichart P, Sidransky D (Eds.): World Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours. IARC Press: Lyon. 2005. ISBN 92 832 2417 5.
[5] Jankowski DS. Traumatic bone cysts in the jaws of a Hong Kong Chinese population. Clinical Clinical Radiology. 1995;50(11):787-91.
[6] Kawamata A, Takai Y, Kanematsu N, Fujiki Y. Bone scintigraphy in solitary (simple) bone cyst of jaw. Oral Radiology. 1993;9(2):01-06.
[7] Cortell-Ballester I, Figueiredo R, Berini-Aytés L, Gay-Escoda C. Traumatic bone cyst: A retrospective study of 21 cases. Med Oral Patol Oral Cir Bucal. 2009;14:E239-43.
[8] Diago M, Bielsa J, Marco J. Surgical treatment and follow-up of solitary bone cyst of the mandible: A report of seven cases. British Journal of Oral and Maxillofacial Surgery. 2001;39(3):221-23.
[9] Strabbing E, Gortzak R, Vinke J, Saridin CP, Merkeysten JP. An atypical presentation of a solitary bone cyst of the mandibular ramus: A case report. Journal of Cranio-Maxillofacial Surgery. 2011;39:145-47.
[10] Hansen LS, Sapone J, Sproat R. Traumatic bone cysts of jaws. Report of sixty six cases. Oral Surgery, Oral Med Oral Pathol. 1974;37(6):899-910.
[11] Forssell K, Forssell H, Happonen RP, Neva M. Simple bone cyst. Review of the literature and analysis of 23 cases. Int J Oral Maxillofac Surg. 1988;17(1):21-24.
[12] Harnet J, Lombard T, Klewansky P, Rieger, Tempe M, Clavert J, et al. Solitary bone cyst of the jaws: A review of the etiopathogenic hypotheses. Journal of Oral and Maxillofacial Surgery. 2008;66(11):2345-48.
[13] Shear M. Cysts of the oral region. 3rd ed. Oxford: Wright PSG; 1992.
[14] Satish K, Padmashree S, Rema J. Traumatic bone cyst of idiopathic origin? A report of two cases. Ethiop J Health Sci. 2014;24:183-87.
[15] Xanthinaki A, Choupis K. Traumatic bone cyst of the mandible of possible iatrogenic origin: A case report and brief review of the literature. Head & Face Medicine. 2006;2(1):40.
[16] Kuttenberger J, Farmand M, Stöss H. Recurrence of a solitary bone cyst of the mandibular condyle in a bone graft. Oral Surgery, Oral Medicine, Oral Pathology. 1992;74(5):550-56.
[17] Brecelj J, Suhodolcan L. Continuous decompression of unicameral bone cyst with cannulated screws: A comparative study. Journal of Pediatric Orthopaedics B. 2007;16(5):367-72.
[18] Dormans J, Sankar W, Moroz L, Erol B. Percutaneous intramedullary decompression, curettage, and grafting with medical-grade calcium sulfate pellets for unicameral bone cysts in children: A new minimally invasive technique. Journal of Pediatric Orthopaedics. 2005;25(6):804-11.
[19] Kumar L, Kurien N, Thaha K. Traumatic bone cyst of mandible. Journal of Maxillofacial and Oral Surgery. 2015;14(2):466-69.
[20] Copete M, Kawamata A, Langlais R. Solitary bone cyst of the jaws: Radiographic review of 44 cases. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 1998;85(2):221-25.
[21] Patil SP, Sodhi SJ, Mishra SS, Farha SS. Solitary bone cyst of posterior maxilla: A rare presentation. Contemp Clin Dent. 2019;10:166-69.
[22] Sánchez AS, Balandrano AGP, Castillejos RD, Ham GC. Maxillary and mandibular solitary bone cyst. Case report and literature review. Revista Odontológica Mexicana. 2018;22(1):39-44.
[23] Brunet-Llobet L, Lahor-Soler E, Mashala EI, Miranda-Rius J. Continuous Surgical Decompression for Solitary Bone Cyst of the Jaw in a Teenage Patient. Case Rep Dent. 2019;2019:9137507.

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