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Case Report

Dentistry Section

A Case of Osseous Choristoma of Submental Region- Cone Beam Computed Tomographic Findings

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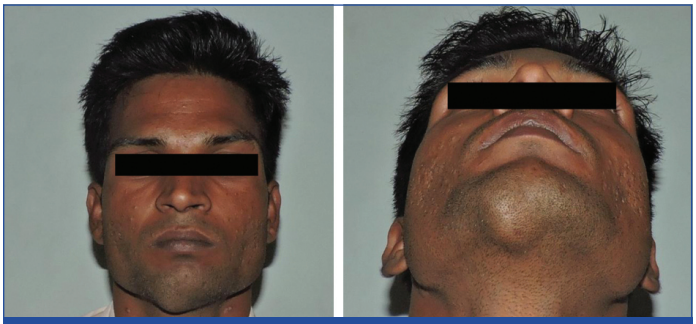
ABSTRACT

Soft tissue radiopacities of head and neck region are intriguing to the oral and maxillofacial radiologist. Osseous choristoma is seldom included in the list of probable differential diagnosis, since its occurrence in the region is a rare phenomenon. Here a case report of osseous choristoma in the submental region of a young male patient has been documanted. This case report highlights the nuances in the radiological interpretation of such radiopaque lesions.

Keywords: Choristoma, Radiopacity, Submental

CASE REPORT

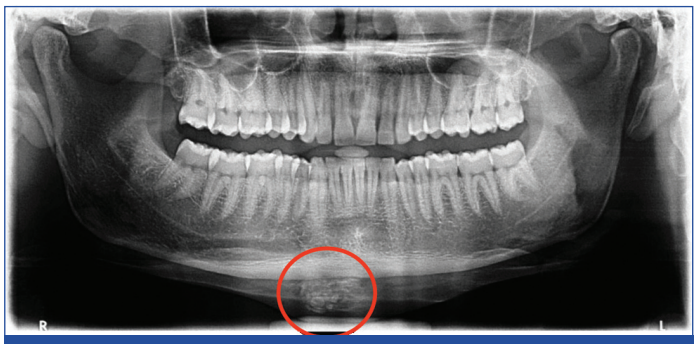
A 38-year-old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling on the lower front 1/3rd region of the face since 3 years. History revealed before 3 years the swelling was smaller in size and increased gradually to its present dimension causing aesthetic concern and discomfort, it was however not painful. There was no associated relevant history. Past medical and dental history were not remarkable. Extra oral examination revealed a unilateral, solitary well-defined swelling seen on the sub-mental region on the right side, which was roughly ovoid in shape approximately measuring 2.5×3 cm in size, extending approximately 0.5 cm lateral to midline and from the inferior border of mandible to about 3 cm into the sub-mental region [Table/Fig-1]. Overlying skin was normal as the surrounding area. On palpation, swelling was non-tender, firm to hard in consistency, slightly movable with well-defined margins.



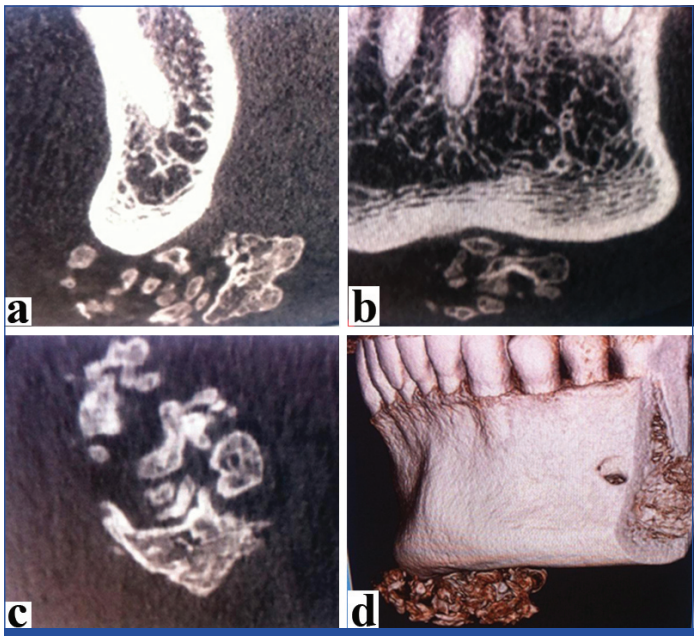
[Table/Fig-1]: Solitary extraoral swelling on right submental region.

Panoramic radiograph [Table/Fig-2] showed multiple clustered elliptical radiopacities in the sub-mental/mid-symphysis region below the lower border of the mandible measuring approx 2×6 mm with ill-defined peripheral borders. Internal structure shows mixed density.

CBCT scan [Table/Fig-3] was done. Coronal, sagittal and axial tomographic images showed multiple radiopaque structure in the soft tissue in relation to the left side of anterior mandible in the region of 31,41 42, 43 and 44. The size of this aggregate was 28 mm antero-posteriorly×22.4 mm mesio-distally×10.4 mm supero-inferiorly. The radiopacities were of varying sizes from very small to large. They were arranged in a whorl like fashion. The largest appears to be a coalescence of smaller mass and measuring approximately 13.9×10.1 mm in dimensions. The individual calcified masses were well-defined, irregular in shape (ranging from ovoid to Chinese letter pattern) and were heterogeneous in their density showing a sclerotic



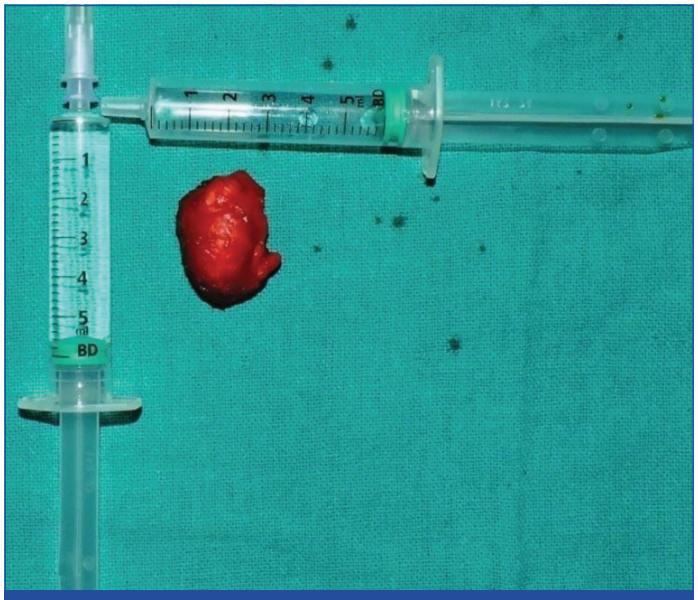
[Table/Fig-2]: Panoramic radiograph showing multiple clustered elliptical radiopacities in the sub-mental/mid-symphysis region below the lower border of the mandible.



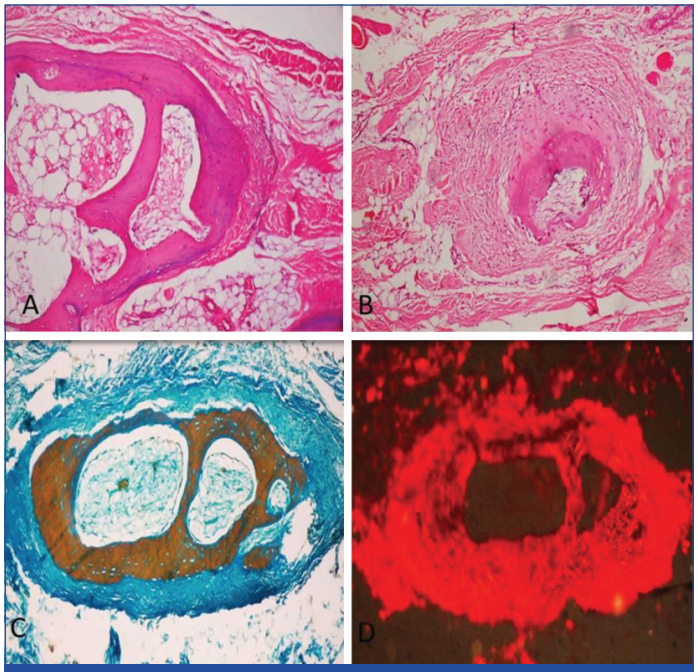
[Table/Fig-3]: a) Sagittal section of CBCT: Individual radiopacity are distinct with an evident corticated margin and internal mixed density. The small radiopacities appear to coalesce to form a large mass.; b) Coronal section of CBCT: The lesion is not in direct contact with the inferior border of mandible; c) Axial section of CBCT showing the conglomerate of radiopacities; d) 3D reconstruction.

margin with amorphous, radiodense internal structure. The lesion was completely free from the cortex of the mandible in the region. Fine Needle Aspiration Cytology (FNAC) was negative. Provisional diagnosis of calcified lymph node was made with a differential diagnosis (d/d) of cysticercosis cutis and dystrophic/metastatic calcification. The patient consented for excision and was referred to Department

of Oral Surgery for further treatment. The lesion was accessed extra-orally through a dermal incision under general anesthesia. Blunt dissection was used to free the lesion from its periphery. The lesion was removed and the soft tissue layers were primarily closed with sutures [Table/Fig-4]. Histopathological examination of the surgical specimen showed mature lamellar bone formation within a fibrofatty connective tissue in the decalcified Haematoxilin and Eosin (H&E) stained section. The lamellated mass of vital bone was surrounded by dense fibrous connective tissue. The bone mass showed peripheral rimming of osteoblast cells with osteocytes present in the osteocystic lacunae. Endothelial cell lined blood vessels, few engorged with RBCs along with hemorrhagic areas also present within connective tissue [Table/Fig-5a,b]. To assess the nature of maturation of calcifications, a panel of special stains including tetrachromic VOF (Verde Luz-orange G-acid fuchsin) and picrosirus red was performed. Tetrachromic VOF shows areas of varied mineralization with central brown and peripheral blue staining suggesting more matured lamellated bone in the center of calcified mass [Table/Fig-5c]. In concordance, picrosirus red stain showed red birefringence which also depicts its mature behavior [Table/Fig-5d]. Based on the radiographic interpretation and histopathological findings, final diagnosis of osseous choristoma in the submental region was made.



[Table/Fig-4]: Surgically excised specimen.



[Table/Fig-5]: a) & b) shows mature lamellar bone formation within fibrofatty connective tissue in H&E stained section (10X); c) Tetrachromic VOF stain shows areas of varied mineralisation with central brown and peripheral blue staining suggesting more mineralised part in the centre; d) Picrosirus red stain shows red birefringence depicting more matured calcification.

DISCUSSION

The term osseous choristoma was introduced by Krolls SO et al., in 1971. Osseous choristoma are uncommon benign lesion that are characterized by ectopic bone pattern in the soft tissue of head and neck region. The pathophysiology accountable for the ossification process in soft tissue is indistinct [1]. The choristoma is a tumourlike mass of normal cells in an abnormal location [2]. It is defined as a development of normal tissue in an abnormal location due to metabolic variations in pluripotent mesenchymal cells as a response to some indefinite stimulus [1,3]. Choristoma rarely occur in the maxillofacial region. It is also referred as soft tissue osteoma, a rare benign lesion of oro-maxillofacial region, while they are most frequently located in the dorsum of the tongue near the circumvallate papillae or foramen caecum. On rare occasions, they can be seen in the buccal mucosa, masseter muscle and submandibular region. The lesions have been shown to be most prevalent in people aged between 20 and 40 years. The majority of patients (72%) were female, with a female/male ratio of 2.7:4 [4]. Since 1913, 97cases of osseous choristomas have been reported in the English literature (PUBMED search). The lesions have a striking predilection for the tongue, accounting for 77 of the 97cases (79%). The occurrence of an osseous choristoma in the buccal mucosa is even more rare, only 14 cases (14%) have been reported so far [1,3] [Table/Fig-6]. The remaining 6 cases were seen in either the lingual aspect of the alveolar process of the anterior mandible or the mandibular buccal vestibule i.e. 4 cases (3%) and 2(1%) at the inferior border of right mandibular angle [5]. To the best of author's knowledge, the present case is the fourth to be reported in the submental region and is of interest due to its rare occurrence in the submental region . It is a challenge to make the diagnosis of osseous choristoma because of its anatomic location, number and pattern of calcification which is similar to various other radiopaque lesion. Cone Beam Computed Tomography (CBCT) has revolutionised head and neck imaging in the recent years. Its role is phenomenal in diagnosing complex lesions. Although CT and MRI have been used to diagnose osseous choristoma, no report of CBCT has been documented till date.

Total No. of documented cases-97	Percentage	Location in the head and neck
77 cases	79%	Tongue
14 cases	14%	Buccal mucosa
4 cases	3%	Mandibular buccal vestibule
2 cases	1%	Inferior border of right mandibular angle
Our case		Right submental region

[Table/Fig-6]: Documented cases of osseous choristoma in the head and neck region.

Soft tissue radiopacities of head and neck are a diagnostic dilemma to the oral radiologist. Osseous choristoma can be easily misdiagnosed as sialoliths, calcified lymph nodes, cysticercosis cutis, myositis ossificans, phleboliths, but these lesions can be distinguished based on their radiographic appearance.

Sialoliths appears as a white or gray opacity, of round to oval or elliptical shape which simulate a whole tooth or root. Surface is smooth or slightly irregular and rough with a laminated calcified mass. Calcified lymph nodes appear as mottled areas of calcific density, which are often multiple and are distributed along the course of cervical, submandibular and digastric node chains. It may be irregular with a cauliflower appearance and smooth contour [6-8]. Cysticercosis cutis appears as a small or slightly elongated as elliptical or ovoid radiopaque masses of 1cm to several mm in diameter. It often resembles grains of rice. Myositis Ossificans appear as a calcifications of muscle often follows a single acute

traumatic episode or a series of minor traumatic episodes which develops in a centrifugal pattern i.e., flocculent at first, becomes more dense, and finally shows the appearance of bone. It appears as a mass with a laminated character. Phleboliths are round or ovoid and varying in diameter from several mms to cms and rarely solitary, many in number from several to dozens within the affected areas. It is arranged as concentric lamellae of calcified material separated by radiolucent bands of uncalcified layers of the thrombus which appear as calcified onion rings [9,10].

As observed by Yoshimura H et al., on immunohistochemical examination, BMPs (Bone morphogenetic protein) were expressed in the osteoblasts surrounding the ectopic bone. Their results indicated that BMP-2 and 4 was associated with the ossification of the osseous choristomas [11].

In the present case, patient's history combined with clinical findings, was supplemented with appropriate imaging modalities and histopathological examination were used to establish a definitive diagnosis. The patient's history bears some critical clues as to the pathological condition presented, including slow growth independent of any salivatory action such as feeding, no evidence of predisposition to local or systemic infections and no history of respiratory problems. Clinical and histopathological findings were in line with osseous choristoma as described in the literature.

CONCLUSION

The oral radiologist plays a crucial role in the diagnosis of soft tissue radiopacities of the head and neck region. The soft tissue osteoma is essentially an osseous tissue found in an abnormal location. The characteristic features of radiopacity itself indicate the nature of the lesion. The marginal cortication with a heterogeneous internal structure separates this entitiy from other probable differential diagnosis. Treatment is usually conservative surgical excision. Recurrence is rare.

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