



Case report

Solitary submandibular soft tissue osteochondroma: A rare case report

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ABSTRACT

Introduction: Soft tissue osteochondromas are rare benign tumors containing bone and cartilage that form in mesenchymal tissues with no connection to adjacent bone, cartilage, or periosteum. They mimic endochondral ossification and are usually encased in a fibrous tissue capsule. The occurrence of the tumor is extremely rare in the maxillofacial region.

Case presentation: A case of soft tissue osteochondroma in the submandibular region of a 47-year-old man with a medical history of muscular dystrophy and schizophrenia is presented here. The tumor had been gradually growing for 11 years before the patient's presentation to our clinics. Radiographic and clinical examination revealed a radiopaque mass in soft tissue that extended from the left anteroinferior border of the mandible and with no connection to the mandibular periosteum. Treatment involved surgical excision. A diagnosis of soft tissue osteochondroma was established by histopathological examination.

Clinical discussion: Previously, there have been only three reported cases of soft tissue osteochondroma in the maxillofacial region. The cause of this tumor is not clearly understood, with multiple hypotheses being proposed. Diagnosis usually involves radiology and histopathology, and the tumor is completely amenable to surgical excision.

Conclusion: Extraskelletal osteochondroma should be considered in the differential diagnosis of hard tissue lesions with no apparent connection to the underlying bone in the maxillofacial region. Although considered rare, clinical awareness about this tumor aids the practitioner in identifying, diagnosing and properly managing this tumor. There has been no report of recurrence or malignant transformation of the lesion.

1. Introduction

Osteochondromas are benign tumors that usually present in the axial skeleton, arising from osseous and cartilaginous tissue. However, a rare variant of this benign tumor may form in the soft tissue with no apparent connection to bone or periosteum. This lesion is termed soft tissue or extraskelletal osteochondroma. Conventional osteochondromas may appear in the maxillofacial region, with reported cases attached by a bony pedicle to the mandibular condyle and coronoid process [1]. Maxillofacial presentation is generally considered atypical. However, based on reported cases, it appears extremely rare for osteochondroma to form in the soft tissues of the maxillofacial or head and neck regions. To the best of our knowledge, there have been only three such reported cases [2–4]. In this report, we present a new case of extra-skelletal

osteochondroma in the submandibular area. This work has been reported in line with the SCARE 2020 criteria [5].

2. Case presentation

A 47-year-old man with a medical history of muscular dystrophy and schizophrenia was referred to the oral maxillofacial surgery clinic at King Saud Dental University Hospital in Riyadh, Saudi Arabia after he had presented to the emergency clinic with a chief complaint of having a protuberant mass that extended from his left chin region. The mass had been gradually extending downward for the past 11 years. He did not seek any medical attention during that time. The patient had a history of multiple falls resulting in fractures to the right femur and humerus and confinement to a wheelchair at the time of presentation. He was also on

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the antipsychotic risperidone (2 mg daily) at presentation.

Clinical examination revealed an extraoral exophytic growth that extended from the left anteroinferior border of the mandible downwards. Upon clinical palpation, the lesion had well-defined borders with a non-tender bony hard consistency which did not seem to be freely mobile. The overlying skin was slightly distended (Fig. 1, A). Cone-beam computed tomography (CBCT) imaging showed a midline soft tissue radiopacity inferior to the chin (Fig. 1, B–D). The radiopacity of bone had an outer cortical layer and inner trabecular bone. The lesion did not seem to be attached to the mandible or any muscle. According to the radiographic presentation, the appearance was suggestive of a soft tissue osteoma of osteochondroma. According to both clinical and radiographic presentation, the consensus of the oral and maxillofacial surgeons (AA, MB, and AFA) was to perform excisional surgery. The patient consented to the procedure and was later admitted to the hospital for surgery under general anesthesia. A skin incision was carried to expose the bony exostosis, which was then bluntly dissected until freed from the underlying tissues (Fig. 2, A–B). The duration of the procedure was 2 h in which blood loss was minimal (<100 cc), and the patient was discharged on the by the end of the day.

Macroscopically, a fungiform $3.5 \times 3 \times 1.5$ cm hard and soft tissue excision was received. The soft tissue part was composed of hairy skin overlying an internal yellowish fatty area. The rest of the specimen was made up of hard bony tissue covered by firm fibrous tissue. Some of the hard tissue was almost at the point of puncturing the skin externally.

Microscopically, the external portion was skin comprising epidermis, dermis and appendages, and subcutaneous fat. The hard tissue showed an orderly arrangement of a fibrous capsule, investing numerous bony trabeculae (many rimmed by plump osteoblasts) with a well-vascularized fibro-fatty marrow. The most internal part continuous with the bony tissue was a cap of cartilage, which was again invested by a dense fibrous connective tissue covering. Generally, the lesion appeared like a miniaturized endochondral ossification with the most matured bone located peripherally (Fig. 2, C–D). Since the whole lesion was invested in soft tissue with no connection to the adjacent mandible, a diagnosis of soft tissue osteochondroma was made.

A weekly follow-up regimen post-surgery was implemented for one month, which was uneventful. The patient is currently on a biannual follow-up protocol one-year post-surgery. Clinical and radiographic examinations showed no signs of recurrence (Fig. 3, A–B).

3. Discussion

Cartilaginous tumors such as osteochondromas represent the majority of benign bone tumors (30%). They usually arise from the cartilaginous cap at the metaphysis and diaphysis of long bones such as the tibia and femur [6]. It is well established that endochondral ossification occurs in long bones instead of intramembranous ossification that is the pathway of development for most bones in the maxillofacial region [1]. Considering this, the presence of a solitary osteochondroma in facial

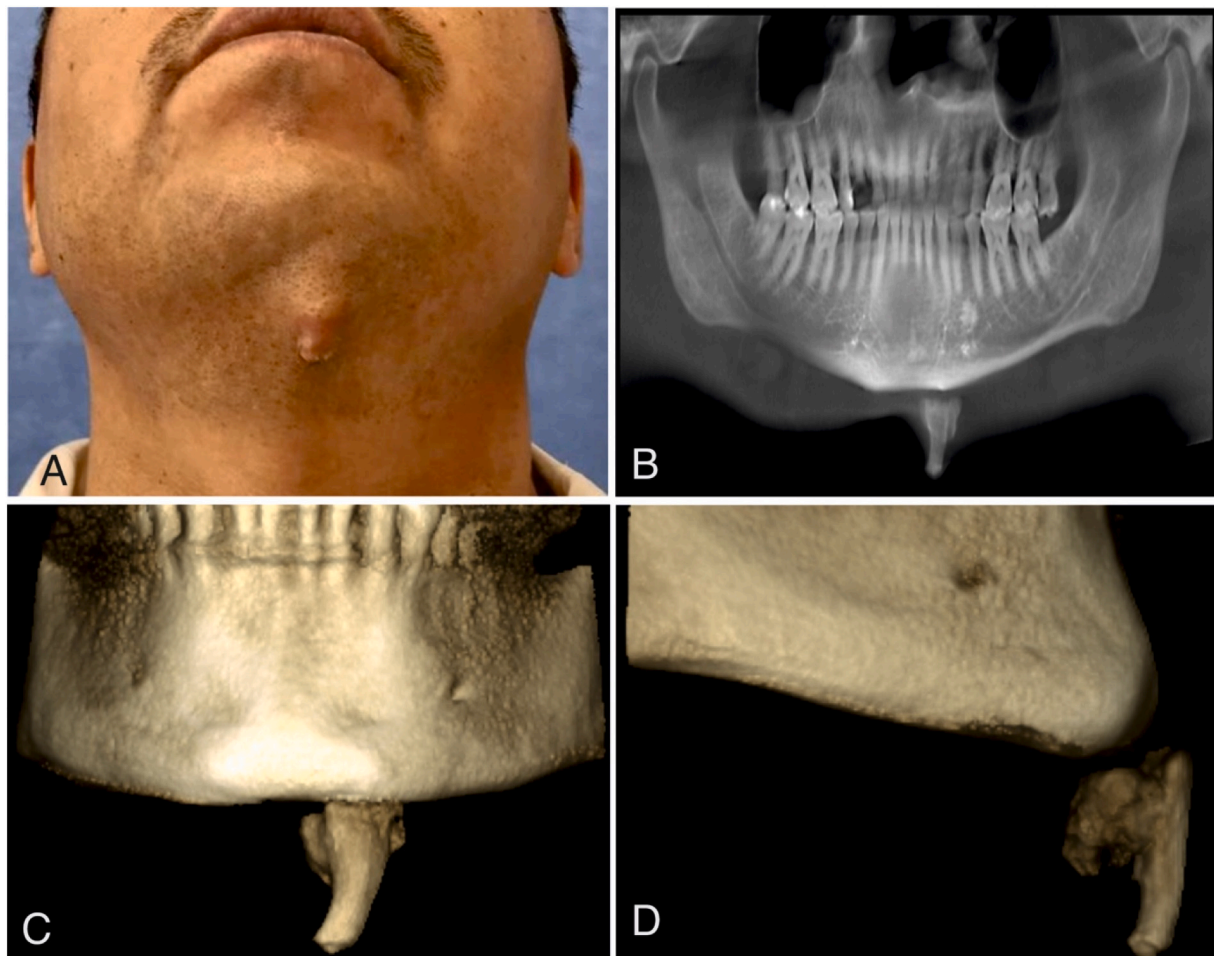


Fig. 1. (A) Clinical photograph showing a protuberant mass just inferior to the chin. The overlying skin is slightly distended with no apparent signs of inflammation or infection; (B) CBCT panoramic image reconstruction shows an exophytic mass at the lower left chin region of the mandible, it has a corticated layer engulfing inner trabecular bone; (C) frontal and (D) lateral views on 3D CBCT image rendering showing how the mass has a flat broad base superiorly and conforms to conical shape as it extends inferiorly. No apparent connection can be seen between the mass and overlying mandible.

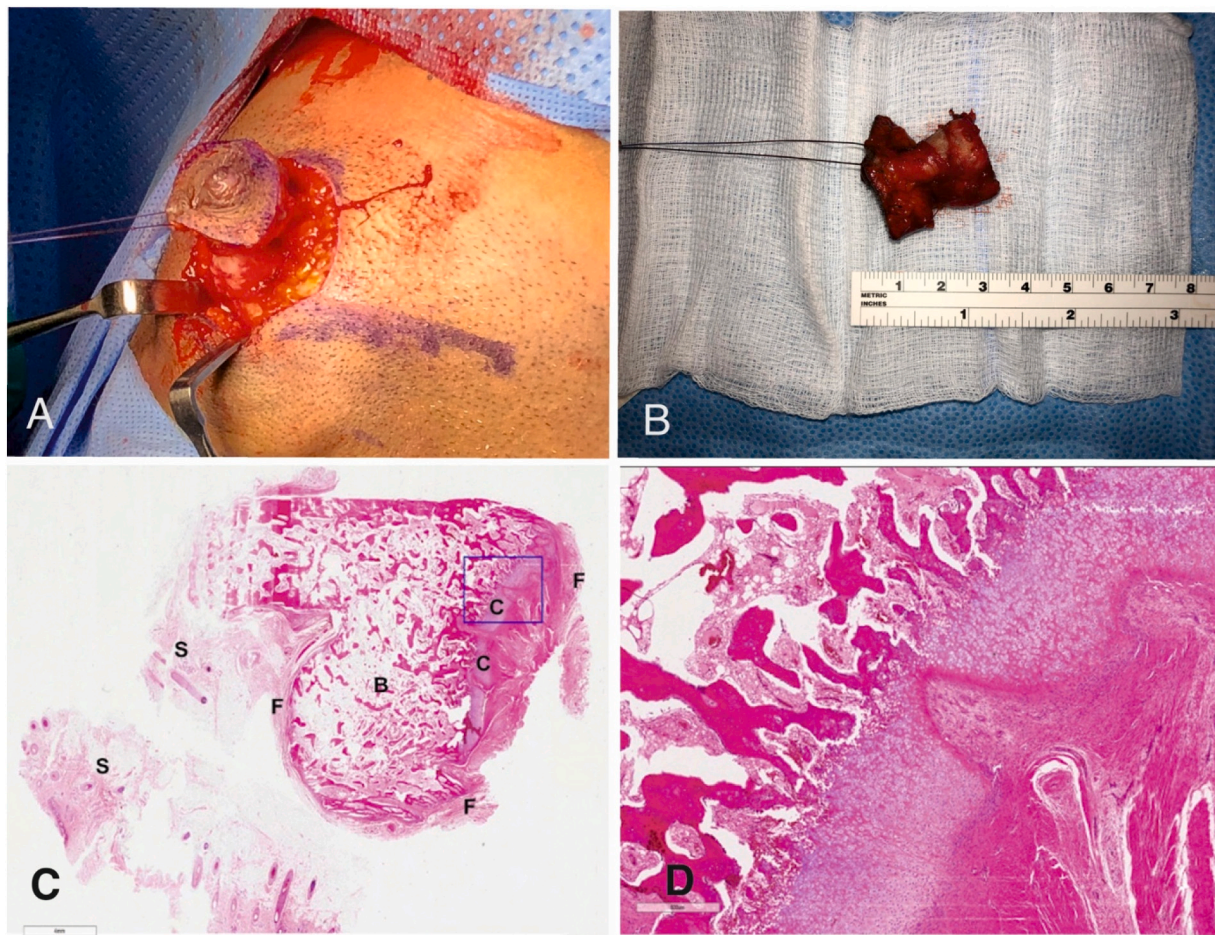


Fig. 2. (A) Intraoperative images showing exposure of the bony mass; (B) mass after excision of the whole lesion; (C) Photomicrograph showing the essential components of the whole lesion; (D) The inset box in C is magnified.

In the whole slide photomicrograph (C), from left to right, the skin appendages (S) overlying the fibrous cap (F) investing the lesion and the bony component (B) immediately subjacent. The deeper part of the lesion closer to the mandible comprised the cartilaginous part (C) covered by a thick scar-like fibrous tissue (F). Some of these features are better appreciated as magnified in (D). Scale bars C: 4 mm; D: 600 μ m.

bones (with features of endochondral ossification) is rather an unusual finding. Such lesions are rare in the head and neck region, with very few cases described. Even rarer are extraskeletal osteochondromas [2–4], as in the present case in which the tumor was present in submandibular soft tissue without attachment to the overlying mandible or surrounding muscle. This unique presentation makes this case worth documentation in the scientific literature.

Soft tissue or extraskeletal osteochondromas are benign, rare occurring tumors that arise within the mesenchymal tissues, not connected through a pedicle with bone or joints, and commonly occurring in hand and feet. Only three previous reports have described soft tissue osteochondroma in the head and neck region. These are summarized in Table 1. Among all benign soft-tissue tumors, cases of extraskeletal osteochondroma account for an estimated 1.5% [7]. Lesions to be considered as a differential diagnosis in well-circumscribed bony masses confined within the soft tissue of the oral and maxillofacial skeleton include tumoral calcinosis, lipomatous lesion, myositis ossificans, extraskeletal osteosarcoma, and synovial sarcoma [3,8–12].

The etiology of soft tissue osteochondroma, however, is not well understood. It remains unknown whether such lesions are developmental, reparative, or neoplastic in origin [1]. Many hypotheses have been proposed as to what causes such tumors to develop [7]. One hypothesis states that extraskeletal osteochondromas develop as a result of the metaplastic activity of mesenchymal fibroblasts in loose connective tissues, while another states that it stems from pluripotent cell lines

delineated from connective tissues, joint synovium, or tenosynovium [13]. Adipose tissue metaplasia is also considered a cause, resulting in the metamorphosis of a lipoma into an extraskeletal osteochondroma [14]. Trauma of repetitive nature is also proposed as a provocative factor in developing extraskeletal osteochondroma [13,15]. In our present case, we speculate that given the patient's history of falls, it may indicate that a possible relationship with trauma exists here. Extraskeletal osteochondromas are most common in patients aged 30–60 years. However, they may also occur at any age. A slowly growing soft-tissue mass accompanied by pain or tenderness are the initial symptoms patients present with the lesion [15]. Histologically, extraskeletal osteochondromas present with a mature center of trabecular bone and a peripheral hyaline cartilage cap contained by a thin fibrous capsule. The demonstration of prominent endochondral ossification within the core areas along with areas of trabecular and lamellar bone [2–4,7] is per our histopathological findings as well.

Soft tissue osteochondromas are usually excised in their entirety as the standard treatment. Recurrence has not been reported [2–4,8]. Clinical awareness of this unusual tumor presentation as a bony lesion in soft tissue with no connection to any adjacent hard tissue is important. No malignant transformation has been reported. CT and MRI help to delineate the extraskeletal origin, and histopathology confirms the diagnosis [3].

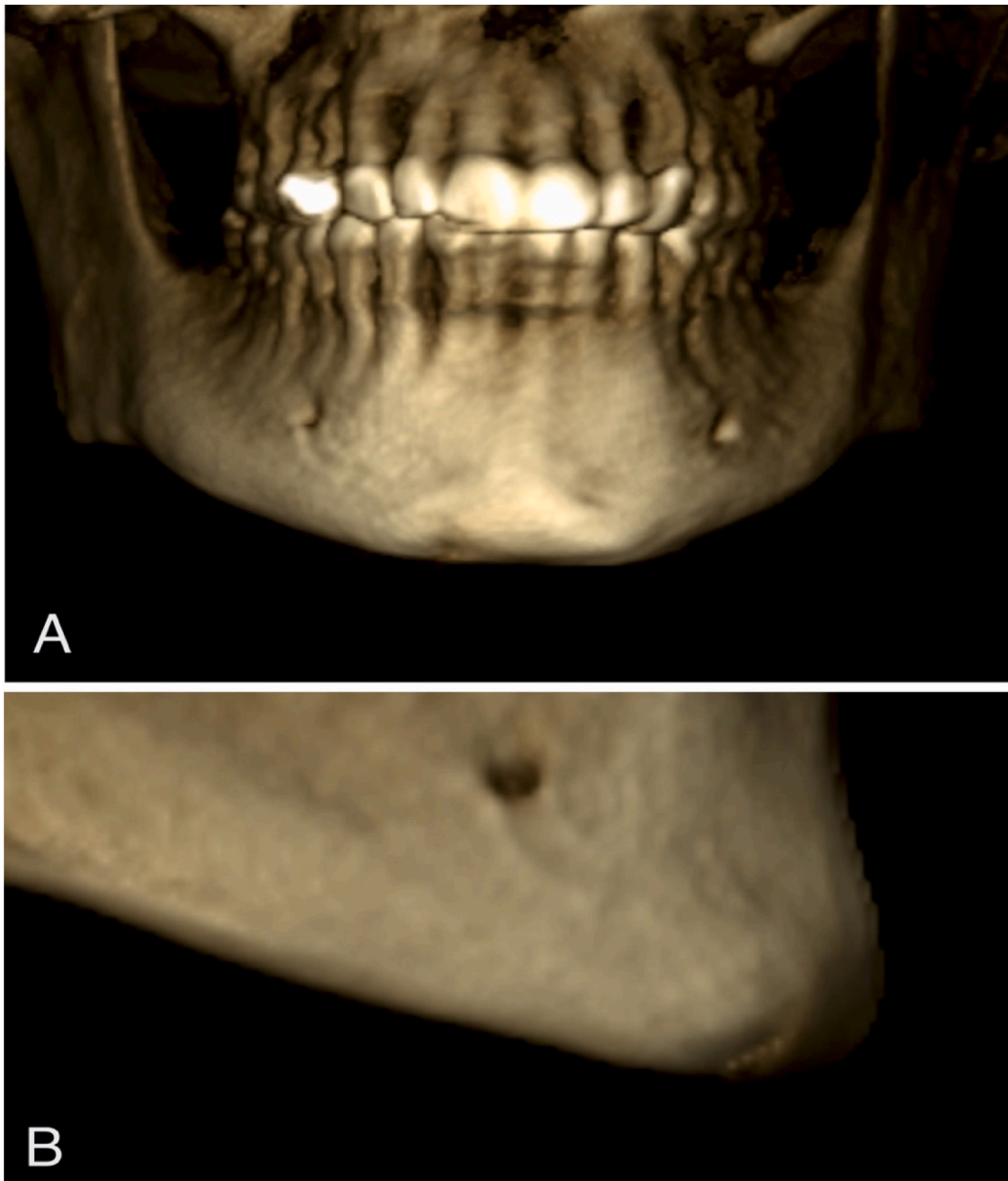


Fig. 3. (A) frontal and lateral; (B) views on 3D CBCT image rendering, showing no signs of tumor recurrence after excision.

4. Conclusion

This report underlines that extraskelatal osteochondroma should also be included in the differential diagnoses of well-circumscribed calcified lesions in the soft tissues of the oral and maxillofacial region. It is unknown what role, if any, was played by the underlying muscular dystrophy in this patient in his presentation with soft tissue osteochondroma.

Ethical approval

IRB approval was waived through the College of Dentistry, King Saud University.

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Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Table 1
Reported soft tissue osteochondromas in the head and neck region.

Author	Age/ sex	Site	Presenting symptoms	Treatment	Recurrence/ follow-up
Sakai et al. [2]	32/F	Mandible angle	Incidental finding	Surgical excision	None/2 years
Singh et al. [3]	42/ M	Nape of neck	Localized pain that increased with motion Limited mouth opening/ respiratory difficulty during sleep	Surgical excision	None/3 years
Cho et al. [4]	2/M	Masticatory space	Mass in the chin	Surgical excision	None/2 years
Present case	47/ M	Submandibular region	Mass in the chin	Surgical excision	None/1 year

M, Male; F, Female.

CRediT authorship contribution statement

All authors contributed significantly and in agreement with the content of the manuscript. All authors participated in data collection and in writing of the manuscript.

Dr. Ibrahim I. Bello (Contributor – Write-up)

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Dr. Moayad Baazeem (Contributor – Case Presentation)

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Declaration of competing interest

None declared.

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