A Rare Case Report of Heterotopic Ossification at Maxilla Anterior

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ABSTRACT

Aim To report a rare case of heterotopic ossification (HO) between the maxillary right central incisor (tooth #11) and lateral incisor in a 47-year-old male patient

Case Report A 47-year-old male patient presented with a painless, progressive-growing swelling in the anterior region involving the maxillary right central and lateral incisors for the past 6 months. The patient's medical history was unremarkable. Intraoral examination revealed periodontal loss with prosthetic restorations in the affected region for over 10 years. Radiographs and cone-beam computed tomography (CBCT) images confirmed the presence of HO between the maxillary teeth.

Discussion HO is a rare condition characterized by the formation of bone in soft tissues. It is most commonly seen after trauma or surgery, but it can also occur spontaneously. The exact cause of HO is unknown, but it is thought to be due to a combination of factors, including inflammation, injury, and genetic predisposition. In this case, the patient's history of trauma may have contributed to the development of HO. The patient reported that he had sustained a blow to the anterior region of his mouth 10 years prior to the onset of the swelling. This trauma may have damaged the periodontal ligament, which may have led to the formation of HO.

Conclusion This case report highlights the importance of considering HO in the differential diagnosis of painless, progressive-growing swellings in the maxillary region, especially in patients with a history of trauma.

Keywords Heterotopic ossification, Lesion, Maxilla, Oral cavity, Osteosarcoma

Introduction

Heterotopic ossification (HO) is defined as the formation of lamellar bone in tissues devoid of native bone. The biological process underpinning heterotopic ossification is associated with the presence of osteoprogenitor cells within the ectopic region (1,2). Heterotopic ossification is categorized into hereditary and acquired forms. The acquired form is frequently observed to manifest following trauma or hip surgery. The prevalence of the hereditary form is reported to be exceedingly low(3).

The majority of HO documented in the literature are connected to orthopedic surgeries, particularly total hip arthroplasty. Cases involving the maxillofacial region are infrequent and typically associated with the facial muscles, although a limited number of reports have described HO occurring in the maxillary sinuses (4).

Heterotopic ossification in the oral region can arise from a variety of factors. Incidents such as direct injury to oral tissues, trauma, or surgical procedures can precipitate its occurrence. Additionally, certain medical conditions, including chronic inflammation, genetic predisposition, and fibrodysplasia ossificans progressiva (FOP), may also contribute to the formation of heterotopic ossification in the oral region (4).

The diagnosis of heterotopic ossification in the oral cavity necessitates a comprehensive examination and thorough evaluation. Common symptoms encompass limited mouth opening, pain, swelling, or palpable bone protrusions. Employing both conventional and advanced imaging techniques aids in delineating the boundaries and localization of the lesion. A definitive diagnosis is achieved through histopathological examination, confirming the presence of ectopic bone formation (4).

Case Report

A 47-year-old male patient underwent examination at a dental clinic for evaluation of a painless, progressively enlarging swelling localized in the anterior region encompassing the maxillary right incisor and right lateral teeth. The patient reported a gradual increase in size over the last few years. Following the initial assessment, the patient was referred to our hospital for further examination and assessment.

Upon reviewing the patient's medical history, no systemic conditions were reported. During the intraoral examination, it was noted that the patient had prosthetic restorations covering the teeth in the affected area for over 10 years, along with evidence of periodontal loss. Notably, a fixed tissue growth was observed between teeth number 11 and 12, situated in the papillary region between the two teeth. The growth exhibited an expansive character, measuring approximately 2 cm in diameter, and demonstrated firm consistency upon palpation. Importantly, there was no observed mobility in the teeth within the relevant area (Figure 1).

No specific abnormalities were identified on the patient's panoramic radiography (Figure 2). However, upon further eval-

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uation using cone beam computed tomography (CBCT), a hyperdense focus with well-defined borders was discernible within the relevant region, specifically within the soft tissue (Figure 3).

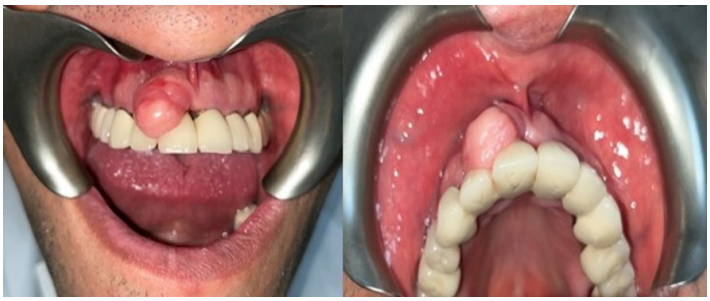

Figure 1: Location of lesion at maxilla anterior.

An excisional biopsy was scheduled for the patient, with a preliminary diagnosis of a local peripheral osseofibroma. The procedure was conducted under local anesthesia, and subsequent histopathological examination was carried out.

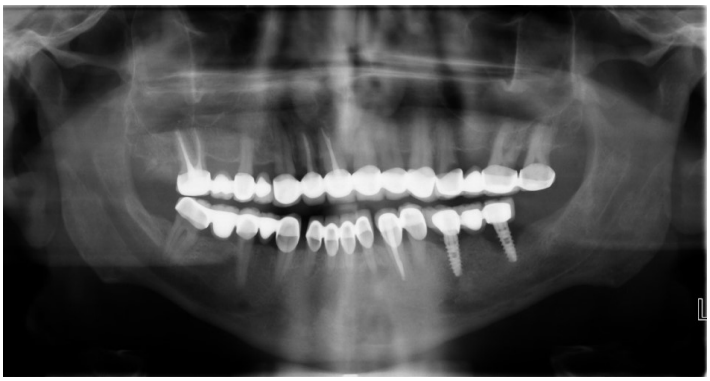

Figure 2: Panoramic radiography of patient.

During the operation, alveoloplasty was performed on the bone tissue in the affected region, along with contour adjustments. The objective was to preserve the aesthetic appearance in the anterior region, achieved by utilizing a connective tissue graft harvested from the palatal region (Figure 4).

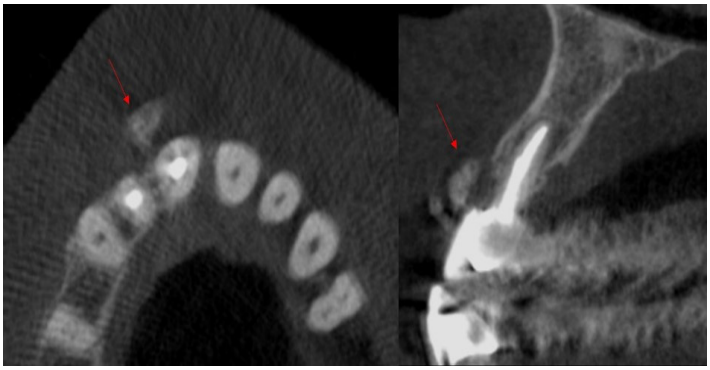

Figure 3: In CBCT axial (left) and sagittal (right) sections, a curved, prominent hyperdense focus (indicated by a red arrow) is observed within the soft tissue, bone density

In the histopathological examination, the evaluated sections revealed a covering of multilayered squamous epithelium over the tissue specimens. Beneath the epithelial tissue, a lesion rich in collagen fibers was observed, composed of triangular or spindle-shaped cells. Notably, collagen fibers exhibited configurations such as swirl-shaped structures and short-course areas that intersected at various points. Additionally, a mineralized region resembling compact bone tissue was identified over a broad area in between. Based on these observations, a diagnosis of 'heterotopic ossification' was established.

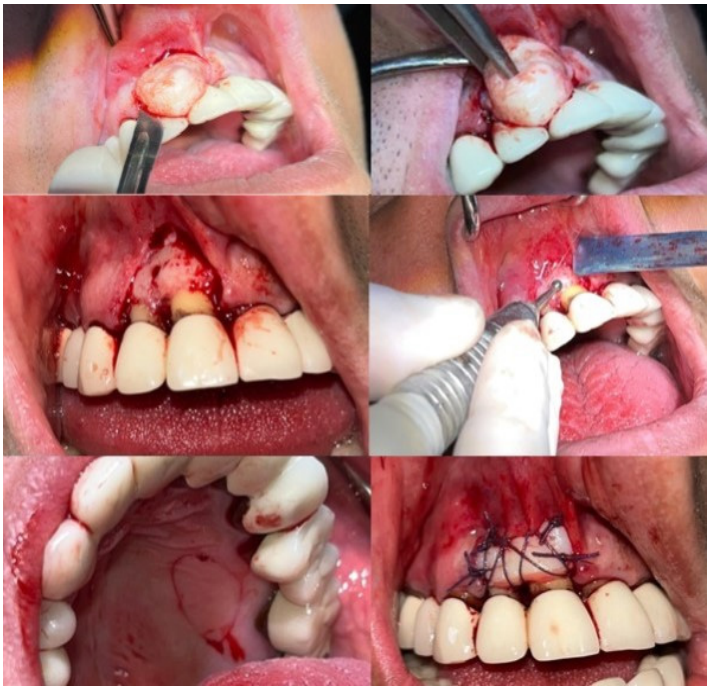

Figure 4: Operation stages: excision, bone arrangement stages, connective tissue removal and wound closure

Discussion

Hyperplastic growths within the maxilla are a commonly encountered phenomenon. These growths typically arise as a response to a stimulus or injury, such as tartar accumulation, ill-fitting dentures, or the presence of foreign materials. Reactive hyperplastic lesions represent non-neoplastic, tumor-like hyperplasias that develop in response to chronic irritation or trauma (5,6). Within this category of common lesions, heterotopic ossification (HO) is infrequently referenced, as evidenced by a limited number of case reports (3,4).

The precise etiology of heterotopic ossification (HO) remains elusive. However, it has been noted that HO is more prevalent in individuals who have undergone significant bone resections or extensive soft tissue dissections (7). The process of HO formation necessitates the presence of osteogenic precursor cells, an inducing substance or event, and an environment conducive to osteogenesis (8).

Radiologically, heterotopic ossification (HO) has been observed to be more prevalent in young males (9). These formations typically manifest as painless, slow-growing, radiopaque lesions, often monitored during routine check-ups (10). For instance, Vencio et al. reported a case of HO situated between the maxillary incisor and lateral incisor in a 13-year-old male patient (11). Hong et al. documented a case of a 3 cm HO, palpable and causing trismus,

located in the masseteric area of the right mandibular ramus region in a patient who sought medical attention due to this issue (12). Additionally, Büyükakyüz et al. reported a case of HO in a patient presenting with complaints of pain in the right maxillary sinus (10).

In addition to its rarity within the maxillofacial region, heterotopic ossifications (HOs) are even more infrequently observed in the anterior maxilla. Potential triggers for its development in the maxilla may include tooth extraction or other irritants. The differential diagnosis of HO necessitates distinguishing it from calcified and ossified structures, ossifying hemangioma, fasciitis ossificans, ossifying pleomorphic adenoma, ossifying fibroma, osteoblastic tumors, and myositis ossificans. Histopathologically, HO typically exhibits a compact bone tissue appearance with inverted lines in decalcified sections (10).

HOs are typically painless and manifest as radiopaque formations observed during routine check-ups. A definitive diagnosis requires histopathological examination (10). Hong et al. reported a case in which they initially clinically and radiologically diagnosed the patient with osteochondroma. Upon surgical intervention, they found the excised mass to be softer than bone and surrounded by cartilage-like tissue. Histological examination revealed normal bone formation covered with osteoids (12).

For the treatment of heterotopic ossification (HO), a follow-up approach is typically recommended rather than invasive interventions, especially if there are no indications of malignancy (13). Güneri et al. have advised surveillance for asymptomatic cases in the maxillary sinus, suggesting a cautious approach without immediate surgery during the initial stage.

However, in specific cases where the condition significantly affects aesthetics and hampers normal lip movements, surgical treatment may be considered, deviating from the typical follow-up approach. In the instance discussed, surgical intervention was chosen due to the disruption of the patient's aesthetics and restricted lip movements.

Conclusion

The rarity of this case and its respective treatment contributes valuable insights to the existing literature. This singular case underscores the importance for maxillofacial surgeons to consider HO, although rare, along with other potential bone lesions like osteoma, osteochondroma, and osteosarcoma, when encountering patients with atypical bone presentations in the maxillofacial region.

Declarations

Author Contributions: *Conception/Design of Study- S.A.T.; Data Acquisition- Z.Y.; Data Analysis/Interpretation- H.A.; Drafting Manuscript- M.M.G.; Critical Revision of Manuscript- M.M.G.; Final Approval and Accountability- M.M.G.; Material and Technical Support- M.M.G.; Supervision- M.R.*

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