Gerodontology

Case report

Reactive osteocartilaginous metaplasia in denture wearers: a rare trauma-related lesion

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Objective: To present a case of reactive osteocartilaginous metaplasia (ROCM) in the anterior edentulous mandibular ridge.
Background: The ROCM secondary to chronic mechanical denture trauma is rare and appears as a focal sometimes painful mass on or near the crest of the edentulous alveolar ridge in long-term denture wearers. The literature review disclosed 24 cases involving more commonly the posterior portion of the mandible.
Materials and methods: An 80-year-old female was referred for the evaluation of a painful, submucosal nodule extending into the vestibular mucosa of the anterior edentulous mandibular region. Microscopically, cartilaginous regions exhibiting sparse hyperchromatic or binucleated chondrocytes transitioned into areas of ossification.
Results: The diagnosis was ROCM. The presence of osteocartilaginous tissue displaying bizarre histopathological features can create a diagnostic dilemma.
Conclusion: Complete conservative surgical excision of this lesion has a very good prognosis. Surgical augmentation of the sharp edentulous mandibular ridges might be needed to avoid continuous irritation and possible recurrence.

Keywords: reactive metaplasia, osteocartilaginous, mandible, bizarre, parosteal proliferation, edentulous alveolar ridge, denture-related lesions

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Introduction

Osseous or cartilaginous/chondromatous changes have been observed microscopically in denture-related fibrous hyperplasias in about 9.3% of the cases[1]. However, the osteocartilaginous tissue formation may be related to the lesion's location, as most cases occurred in the anterior maxillary region, a site known for the presence of cartilaginous embryonic remnants[2]. The development of osteocartilaginous tissue as a phenomenon of reactive metaplasia secondary to chronic mechanical denture trauma is rare. The literature review disclosed 24 cases involving more frequently the posterior followed by the anterior (9/24, 37.5%) part of the mandible[2,3]. This benign condition occurs in long-term denture wearers with atrophic ridges and clinically appears as a focal sometimes painful mass on or near the crest[3,4].

The denture-related reactive osteocartilaginous metaplasia (ROCM) may show atypical histopathological features exhibiting hypercellularity, multinucleate lacunae, cellular pleomorphism and nuclear hyperchromasia of the chondrocytes[3]. The presence of irregular osteocartilaginous tissue displaying bizarre features, suggestive of malignancy, may create diagnostic dilemma, on whether such a lesion is reactive or neoplastic in nature leading to a possible misdiagnosis. We present a case of ROCM in the anterior edentulous mandibular ridge and discuss the lesion's differential diagnosis.

Case presentation

An 80-year-old female visited her dentist complaining of pain and diffuse swelling of the sulcus in the anterior mandibular region respective to an ill-fitting complete lower denture. The patient had been a complete denture wearer for the last 20 years. Her medical history included vascular hypertension and controlled insulin-dependent diabetes mellitus. A periapical radiograph revealed residual root fragments of the mandibular right central and lateral incisors, which removed by the dentist, while the patient was put under antibiotics. The pain improved quickly, but on re-examination 4 weeks later, a nodular lesion remained, and therefore, the patient was referred to us for evaluation and treatment. On intraoral examination, a firm, well-demarcated, submucosal sessile nodule 2 cm in diameter was present on the atrophic edentulous alveolar ridge extending into the vestibular mucosa of the anterior mandibular region respective to the incisors area. The overlying red in colour mucosa showed irregular surface with a fistula opening. A mass exhibiting irregular radiopaque calcifications was localised with introduction of gutta-percha through the fistula; early bone healing was evident at the extraction sites (Fig. 1). The differential diagnosis included fistular granuloma, ossifying fibroma and reactive mandibular osteocartilaginous metaplasia. Under antibiotic prophylaxis owing to the diabetic history, the clearly separated from the underlying cortical bone lesion was excised totally together with healthy surrounding tissue, under local anaesthesia. Histopathological examination disclosed cartilaginous regions surrounded by cellular focally myxoid connective tissue. The hyaline cartilage demonstrating sparse large, hyperchromatic or binucleated chondrocytes (Fig. 2a) transitioned into areas of ossification (Fig. 2b), but there was no evidence of osteoid formation by atypical osteoblasts. The final diagnosis was ROCM of the alveolar ridge. The postoperative course was uneventful; because of the patient's age and medical history, it was decided not to restore the atrophic mandibular bone. The patient is being followed up regularly, and there is no clinical evidence of recurrence after 24 months.

Figure 1 Intraoral radiograph showing irregular calcifications.

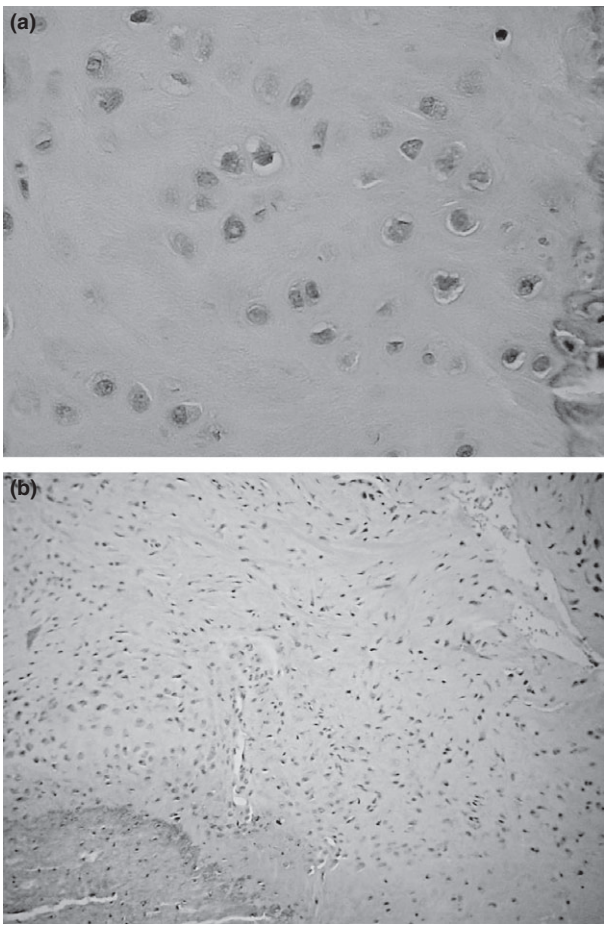

Figure 2 (a) Histopathological features of cartilaginous metaplasia showing few large, hyperchromatic or binucleated chondrocytes. (b) The cartilaginous regions surrounded by cellular focally myxoid connective tissue. The cartilage transitioning into endochondral ossification demonstrated characteristic 'blue staining' with haematoxylin at the interface of cartilage and bone. Haematoxylin and eosin staining; magnification (a) ×400 and (b) ×100.

Discussion

The ROCM as a periosteal response in the edentulous mandibular alveolar ridge is rare. Alternatively, it may be under-reported, because many patients, after provision of dentures, miss their regular follow-ups and present to their dentist only in case of emergency (pain, swelling). Clinically, it presents as a localised enlargement or nodular lesion more frequently in middle aged to elderly female long-term denture wearers, who usually have atrophic alveolar ridge with a sharp crest. The ROCM arises beneath the thin tightly bound mucosa, and it may be associated with pain. Radiographically, focal bone saucerization has been reported in some cases, but no bone destruction[3].

The pathogenesis of the ROCM has not been fully elucidated. The distinction between reactive hyperplasia of cartilaginous remnants and reactive cartilaginous metaplasia was pointed out by Daley *et al.*[3] and Neville *et al.*[4] The hyperplasia may be the result of the reactive proliferation of possible pre-existing embryonic cartilaginous remnants, such as in the maxillary incisive papilla region. The pathogenesis of the cases reported by Stimson[5], Lello and Makek[6] and Magnusson *et al.*[7] may fit into this category. In contrast, in metaplasia, mechanical stimulation and signalling proteins (mainly bone morphogenetic proteins) from the site of injury may induce uncommitted periosteal mesenchymal cells towards chondroblastic or/and osteoblastic differentiation[3,4].

Reactive lesions involving hands and feet[8], long bones, skull[9], maxilla[10] and zygoma[11] may exhibit similar clinical and histopathological features with the denture-related ROCM. However, these lesions, referred in the literature as bizarre parosteal osteochondromatous proliferation (Nora's lesion), may not always be associated with history of trauma or inflammation, and additionally show a tendency for recurrence ranged between 20% and 55%[8,9]. In our case, the osteocartilaginous metaplasia may be related to a possible combination of local and systemic contributing factors. The denture mechanical stimulation, diabetic history predisposing to inflammation and the presence of root fragments in the region may all play a role in the pathogenesis of this particular case; heterotopic bone and cartilage formation induced by implants of mineralised hard tissues have been showed in experimental studies[12].

The main histopathological feature of the ROCM is the irregular aggregates of hyaline cartilage forming a cap at the periphery of the lesion because of fibrous tissue chondroid metaplasia. The hypercellular cartilage lobules exhibiting large, sometimes hyperchromatic or binucleated chondrocytes are separated by spindle cells, but cellular atypia and atypical mitotic figures are not present differentiating this benign lesion from chondrosarcoma and chondroblastic osteosarcoma. An additional characteristic feature of ROCM is the deep basophilia of the new bone formation by endochondral or metaplastic ossification[3,4]. These features are demonstrated in the present case. Benign cartilaginous tumours, such as osteochondroma, is also included in the differential diagnosis, but this rare tumour tend to occur in the temporomandibular joint region, because of the presence of cartilaginous tissue at the mandibular condyle head. Osteochondroma shows in continuity with the underlying cortical bone, whereas the ROCM grows as a mass into the paraosseous soft tissues leaving the cortex of the adjacent bone undisturbed[3,4].

Complete conservative surgical excision of the lesion has a very good prognosis. Recurrences have not been reported after long follow-up periods[3]. Improvement of the sharp alveolar ridges with different augmentation techniques may be indicated to avoid the continuous irritation and possible recurrence. Moreover, the importance of regular follow-up of the denture wearer patients with severely atrophic mandible should be emphasised, because this appears to be an important risk factor for developing ROCM.

Conflict of interest

We declare that we have no proprietary, financial, professional or other personal interest of any nature or kind in any product, service and/or company that could be construed as influencing the position presented in, or the review of, the manuscript.

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