ORIGINAL ARTICLE

Unusual bone-forming lesion affecting complete denture wearer: two case reports

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Introduction: The use of complete removable dentures is associated with a high frequency

Abstract:

of oral mucosal lesions. Among them, denture-induced fibrous inflammatory hyperplasia is one of the most common. Interestingly, some oral lesions occurring beneath complete dentures can present exuberant new bone formation, creating diagnostic difficulties. Patients: We present two interesting cases affecting complete denture wearers, showing each one unusual bone-forming lesion. The first was diagnosed as extensive mature bone formation with osteochondromatous metaplasia foci and the second as an exuberant exostosis. Discussion: Oral soft tissue lesions caused by ill-fitting dentures may present distinct microscopic variations such as osseous and chondromatous metaplasia arising of the connective tissue. These cases evidence that contributing factors for exostosis may be the chronic oral mucosa irritation at the tissue/denture base interface and a response to stresses transferred to the alveolar bone by the ill-fitting denture. Conclusion: Awareness of these lesions is essential to avoid unnecessary procedures, prevent misdiagnosis, reduce patient anxiety, consequently influencing or al health related-quality of life of edentulous patients.

Keywords: Exostoses, Osteochondroma, Metaplasia, Denture, Complete

1. INTRODUCTION

Denture satisfaction is the strongest predictor of oral health related-quality of life as previously demonstrated in edentulous patients wearing removable dentures^{1,2}. However, the use of complete removable dentures is associated with a high frequency of oral mucosal lesions, such as fibrous inflammatory hyperplasia, erythematous candidiasis, traumatic ulcer, angular cheilitis and flabby ridge. These lesions occur frequently in females, with denture use for 16 to 20 years³. Denture-induced fibrous inflammatory hyperplasia is one of the most frequent lesions, commonly associated to an ill-fitting denture^{3,4}. However, some oral soft tissue lesions affecting complete denture wearers may present distinct microscopic variations such as osseous, chondromatous, oncocytic and squamous metaplasia⁴.

Tori and exostoses are osseous protuberances, whose classification depends on anatomic location. The histologic features of tori and exostoses are similar, consisting of mature cortical and trabecular bone. To the best of our knowledge, there are only two case reports showing an unusually large palatal exostosis^{5,6}.

Here, we present two complete denture wearers, each one presenting unusual bone-forming lesion.

2. CASE REPORT

Case 1

A 50-year-old woman was referred to assess a nodular lesion located in the mandible. The patient was user of upper and lower complete dentures 30 years ago, and she reported that the lesion was present about 1 year ago. The medical history was noncontributory and the results of laboratory tests were normal. The oral examination revealed a nontender nodular lesion on the edentulous alveolar ridge of the mandible occurring around the borders of an ill-fitting denture. A panoramic radiograph examination evidenced an irregular radiopaque lesion just above the upper border of the lower alveolar ridge (Fig. 1). Despite this, regarding the clinicopathological features, a reactive/benign proliferative process consistent with denture-induced fibrous inflammatory hyperplasia was favored. During the surgical treatment, it was observed a mass with a hard consistency at the base of the lesion; nevertheless, it was possible to separate from the alveolar ridge. The microscopical examination showed trabecular bone composed by mature lamellar bone and surrounded by connective tissue. The medullary spaces contained loose fibrovascular tissue admixed with adipose marrow and



Figure. 1. Panoramic radiograph showing an irregular radiopaque lesion above of the lower alveolar ridge (arrows).

foci of hematopoiesis (Fig. 2A). At a deeper level, it was visualized focally hyaline cartilage showing calcification and squamous epithelial island in close association with a neurovascular plexus (Fig. 2B). A diagnosis of extensive mature bone formation and osteochondromatous metaplasia foci associated with denture-induced fibrous hyperplasia was made. After 1-year of follow-up, no alteration or recurrence was observed.

Case 2

A 74-year-old man, experiencing problems with his complete dentures, was referred to assess an asymptomatic lesion in the maxilla. During the anamnesis the patient reported that the lesion appeared 56 years ago. The medical history revealed that the patient had arterial hypertension, diabetes mellitus and high cholesterol levels. The intraoral examination showed a large nodular mass, measuring approximately 3 cm x 2 cm, located on the right side of the maxillary posterior edentulous alveolar ridge (Figs. 3A and B). A panoramic radiograph revealed a well-defined radiopaque lesion in continuity with the maxilla, and the computed tomography (CT) scan exhibited a discretely pedunculated lesion showing mixed areas and delimited by hyperdense lines. The 3D reconstruction clearly showed a pedunculated lesion of irregular surface (Fig. 4). The blood count, biochemical tests (serum calcium, phosphorus and alkaline phosphatase), parathyroid hormone (PTH) and urine routine test showed normal values. The lesion was completely removed and the surgical specimen exhibited ample areas of mature compact bone delimiting irregular medullary spaces (Fig. 5). A final diagnosis of exuberant exostosis was favored. After 2-year of follow-up, there is no sign of alteration or recurrence (Fig. 3C).



Figure. 2. The histopathological analysis showed large area of mature lamellar bone surrounded by highly cellular connective tissue (A, x25). Moreover, notice medullary spaces containing foci of erythropoiesis (A, inset). At the periphery, abundant fibrous connective tissue admixed with hyaline cartilage showing calcification (thin arrow) and squamous epithelial island (thick arrow) were observed (B, x25) (H&E stain).

3. DISCUSSION

Interestingly, some oral soft tissue lesions caused by ill-fitting complete dentures present distinct microscopic variations such as osseous and chondromatous metaplasia arising of the connective tissue, as well as oncocytic and squamous metaplasia arising of the residual minor salivary glands⁴. As commented above, unusual self-limiting osteochondromatous metaplasia (Cutright lesion) can be found in the soft tissue beneath ill-fitting complete dentures, especially in the anterior region of the jaws⁴. Cutright lesion is characterized histologically by diffuse deposits of calcium and bone formation, as well as scattered single or clustered cartilaginous foci arranged in various stages of maturation. However, unlike most Cutright lesions reported so far, in the current case 1 extensive mature bone formation and osteochondromatous metaplasia foci at the periphery of the surgical specimen were visualized. These findings have led us to think that Cutright lesion probably represents an early stage of bone and cartilage formations on edentulous alveolar ridge in wearers of complete dentures.

Other interesting finding in the current case 1, inside mature osseous tissue, was the presence of bone marrow

containing characteristic hemopoietic cells such as red cell series (Fig. 2A, inset). Extramedullary hematopoiesis (EMH) is defined as production of myeloid, erythroid and megakaryocytic elements at ectopic sites. The most frequent sites of EMH are spleen, liver, and lymph nodes, but almost every other organ can be involved. It has been frequently described in hematologic diseases such as anemia and chronic myeloproliferative disorders7. To the best of our knowledge, this is first case of EMH affecting a complete denture wearer, which was clinically healthy. However, probable compensatory phenomena must be considered because the patient's age. Furthermore, near the osteochondromatous metaplasia focus it was evidenced a squamous epithelial island in close association with a neurovascular plexus. Epithelial islands located within the jaw bones and gingival tissues are not uncommon and usually represent odontogenic epithelial remnants often associated with neural structures.

Osseous choristoma, which is composed of normal lamellar bone occurring in an abnormal location, should be differentiated from the case 1. Osseous choristomas affecting the oral mucosa are rare and occur preferentially on the tongue, followed by buccal mucosa, palate and gingiva. These oral lesions are generally covered by integral mucosa and it can occur at any age. It has been suggested that osseous choristomas have developmental or traumatic origins. The treatment of choice is the surgical excision, and a definite diagnosis can only be determined after histologic examination⁸. The clinicopathological features from current case 1 precluded the diagnosis of osseous choristoma. In fact, this denture-induced lesion depicted by microscopical analysis periosteum-like tissue surrounding mature trabecular bone, in addition to large areas of reactive fibrous connective tissue and osteochondromatous metaplasia foci.

In the current case 2, the main differential diagnoses were exostosis and osteoma. Exostoses and tori are nodular protuberances of mature bone, whose accurate diagnosis depends mainly on the anatomic location. Torus palatine



Figure. 3. Intraoral view showing adaptation of the prosthesis on the anterior surface of the lesion (A). Extensive nodular lesion on the right posterior alveolar ridge (B). Clinical view after 2-year follow-up (C).



Figure 4. Panoramic radiograph shows a lesion with variable radiopacity in close contact with the maxillary sinus floor (A). Coronal CT shows a well-defined rounded lesion without involvement of the maxillary sinus (B). 3D reconstruction showing a pedunculated lesion (C).



Figure 5. The histopathological analysis revealed a formation of mature compact bone delimiting irregular medullary spaces (A, x25; B, x100) (H&E stain).

occurs along the midline of the hard palate, while torus mandibularis is located on the lingual aspect of the mandible. Buccal and palatal exostoses occur less frequently than tori, and along the buccal aspect of the maxilla or mandible and on the palatal aspect of the maxilla, respectively. The microscopical features of these lesions are similar, consisting of mature cortical and trabecular bone^{5,6}. Interestingly, osteoma, a relatively rare benign osteogenic neoplasm, also shows similar histopathological features⁶. Thus, exostosis and osteoma are distinguished on the basis of their clinical and imaginological characteristics. In fact, such as suggested by Kaplan et al.9, the osteoma diagnosis requires clinical evidence of growth, expansion and/or displacement of teeth. In the current case 2, the patient reported that the maxillary lesion appeared 56 years ago, asymptomatic, with no recent evidence of cortical expansion, supporting a reactive nature. Thus, a diagnosis of exuberant exostosis was favored. To the best of our knowledge, there are only two case reports each one showing an unusually large palatal exostosis5,6. Take together, these cases place in evidence that contributing factors for exostosis may be the chronic oral mucosa irritation at the tissue/denture base interface and a response to stresses transferred to the alveolar bone by the ill-fitting denture.

4. CONCLUSION

In summary, we reported two unusual boneforming lesions affecting complete denture wearers. The first lesion showed extensive mature bone formation with hematopoiesis and osteochondromatous metaplasia, and the second lesion was an exuberant exostosis. A detailed analysis of clinical, imaginological e histopathological features is fundamental for to avoid unnecessary procedures, prevent misdiagnosis and reduce patient anxiety with impact on oral health related-quality of life of edentulous patients.

Conflicts of interest

The authors have no conflict of interest in the present manuscript.

REFERENCES

- 1. Yen YY, Lee HE, Wu YM, Lan SJ, Wang WC, Du JK, et al. Impact of removable dentures on oral health-related quality of life among elderly adults in Taiwan. BMC Oral Health 2015; 15: 1.
- 2. Viola AP, Takamiya AS, Monteiro DR, Barbosa DB. Oral health-related quality of life and satisfaction before and after treatment with complete dentures in a Dental School in Brazil. J Prosthodont Res 2013; 57(1): 36-41.
- 3. Carvalho Mde V, Iglesias DP, do Nascimento GJ, Sobral AP. Epidemiological study of 534 biopsies of oral mucosal lesions in elderly Brazilian patients. Gerodontology 2011; 28: 111-115.
- 4. Cutright DE. Osseous and chondromatous metaplasia caused by dentures. Oral Surg Oral Med Oral Pathol 1972; 34: 625-633.
- 5. Blakemore JR, Eller DJ, Tomaro AJ. Maxillary exostoses. Surgical management of an unusual case. Oral Surg Oral Med Oral Pathol 1975; 40: 200-204.
- 6. Raldi FV, Nascimento RD, Sá-Lima JR, Tsuda CA, Moraes MB. Excision of an atypical case of palatal bone exostosis: a case report. J Oral Sci 2008; 50: 229-231.
- Santonja C, de Sus J, Moragón M. Extramedullary hematopoiesis within endothelial papillary hyperplasia (Masson's pseudoangiosarcoma) of the tongue. Med Oral Patol Oral Cir Bucal 2007; 12(8): E556-559.
- 8. Gaitán-Cepeda LA, Quezada-Rivera D, Ruíz-Rodríguez R. Osseous choristoma of the oral soft tissue. Case report. Med Oral 2003; 8(3): 220-223.
- 9. Kaplan I, Nicolaou Z, Hatuel D, Calderon S. Solitary central osteoma of the jaws: a diagnostic dilemma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008; 106: e22-29.