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Peripheral Osteoma of the Maxillary Alveolar Process

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Abstract: Osteoma is a benign, slow-growing tumor characterized by proliferation of compact or cancellous bone. Solitary osteomas are classified as peripheral, central, or extraskeletal. Peripheral osteomas of the oral cavity are unusual and the maxilla is rarely affected. They manifest as asymptomatic, fixed tumors of bony-hard consistency that may be sessile or pedunculated. Radiographically, a well-circumscribed round or oval radiopaque mass is seen that is microscopically composed of cancellous or trabecular bone. A case of a compact, peripheral osteoma arising from the buccal plate of the alveolar ridge of the maxilla in a 64-year-old patient is presented. According to our literature review, this is the seventh case reported in the maxillary ridge.

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Key Words: Osteoma, peripheral, bone tumors, maxilla, maxillary tumors

Osteoma is a benign, slow-growing tumor characterized by proliferation of compact or cancellous bone.[1,2] Solitary osteomas are classified as peripheral, central, or extraskeletal.[3] Peripheral and central osteomas arise from the periosteum or the endosteum, respectively. Extraskeletal osteomas develop within soft tissue, usually striated muscles, and are considered choristomas. Osteomas may be solitary or multiple, the latter mainly associated with Gardner's syndrome.[2–5]

Peripheral osteomas are found almost exclusively in the skull and maxillofacial bones. The most common site is the frontal sinus followed by the ethmoidal and maxillary sinuses. Osteomas of the oral cavity are unusual and the maxilla is rarely affected.[5–7] There is no age or sex predilection.[2,5] Clinically, they manifest as fixed tumors of bony-hard consistency that may be sessile or pedunculated. Because osteomas are usually asymptomatic, most patients present because of aesthetic considerations such as facial asymmetry or malocclusion. Pain, gagging, nausea, and dysphagia are rarely reported.[4,5]

Radiographically, the lesion appears as a well-circumscribed round or oval radiopaque mass.[3,8,9] Histologically, an osteoma may be composed of dense cortical bone (compact or ivory osteoma) or trabecular, medullary bone (cancellous, trabecular, or spongy osteoma).[5] When fibrous connective tissue dominates the microscopic picture, the lesion is called fibroosteoma or fibrous osteoma.[10]

A rare case of compact, peripheral osteoma arising from the buccal plate of the alveolar ridge of the maxilla in a 64-year-old patient is presented.

Clinical Report

A 64-year-old white woman was referred by her periodontologist for evaluation of a painless swelling on the buccal plate of the right maxillary alveolar ridge. According to the patient, the lesion had been present for several months, and there was no history of trauma or inflammation to the area. Her medical history was noncontributory.

Oral examination revealed a circumscribed tumor on the alveolar maxillary ridge, buccally to the missing second premolar tooth (Fig 1). The

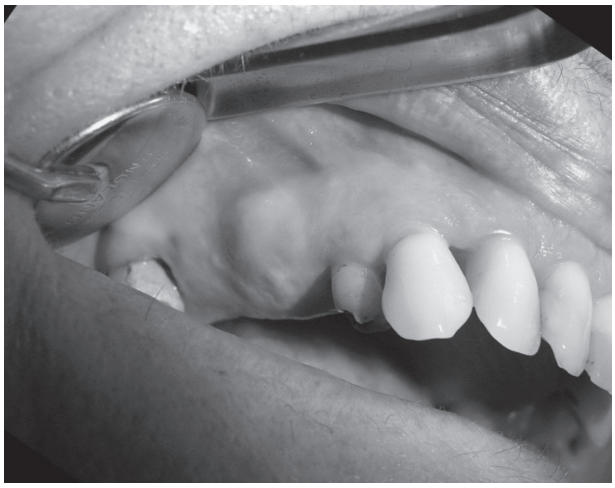

Fig 1 Circumscribed tumor on the alveolar maxillary ridge, buccally to the missing maxillary second premolar tooth.

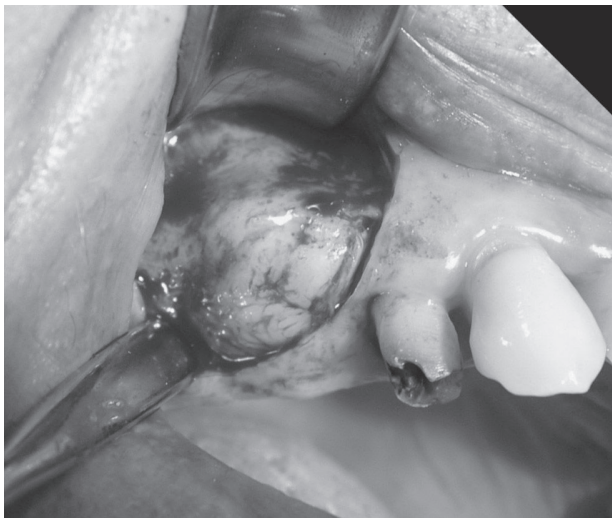

Fig 3 Intraoperative aspect of the lesion.

lesion was approximately 2 cm in diameter. It was covered by normal mucosa, was pedunculated, and on palpation bony-hard and nontender. The maxillary right first molar was also missing and there was a fixed metal–ceramic bridge extending between the first premolar and the second molar. A panoramic radiograph showed a dense, radiopaque mass with ill-defined margins laterally, close to the floor of the right maxillary sinus, and well-defined margins inferiorly to the alveolar ridge (Fig 2).

With the provisional diagnosis of a peripheral osteoma, a full-thickness buccal trapezoidal mucoperiosteal flap was reflected under local anesthesia, and the tumor was exposed (Fig 3). The protruding part of the lesion was removed in toto with an osteotomy on grossly healthy margins and fixed in buffered formalin. The osteotomy margins were carefully smoothed, hemostasis was achieved with packing of collagen sponges, and the surgical site

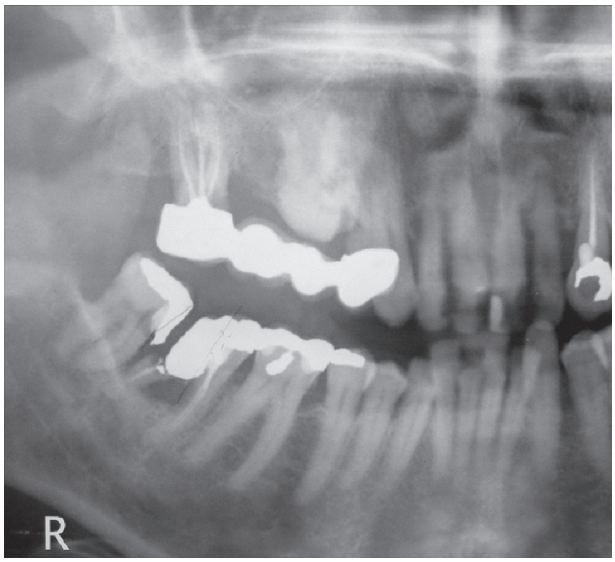

Fig 2 The panoramic radiograph shows a dense radiopaque mass.

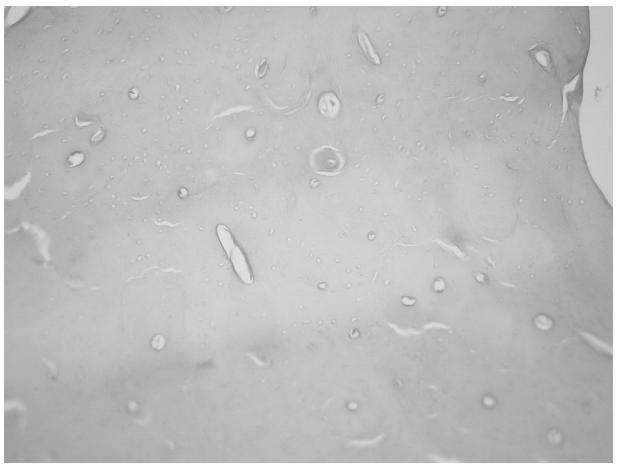

Fig 4 Microscopic examination shows dense, mature cellular bone with well-defined haversian systems (hematoxylin and eosin stain, original magnification ×200).

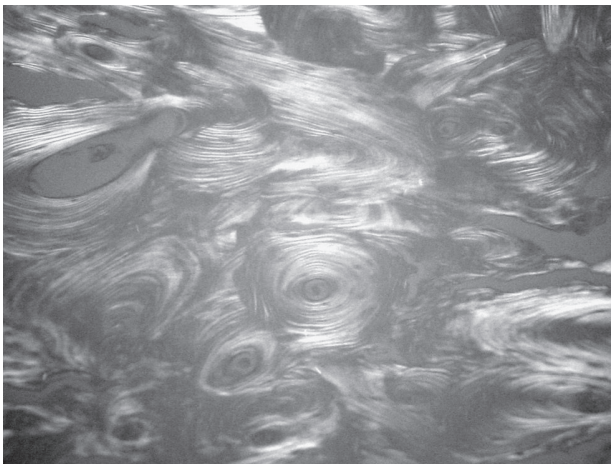

Fig 5 The haversianstems are more evident under polarized light (hematoxylin and eosin stain, original magnification ×200).

was closed primarily. The postoperative course of the patient was uneventful.

Macroscopically, a hard, white specimen measuring 2 × 1 × 1.7 cm was seen. After decalcification, 5-μm thick paraffin-embedded sections were stained with hematoxylin and eosin. The specimen consisted of dense, mature cellular bone with well-defined haversian systems (Fig 4) that were more evident under polarized light (Fig 5). The histologic diagnosis was consistent with an osteoma.

Two years after the excision, the patient was free of signs of recurrence (Fig 6) and the radiographic examination showed focal bone regeneration (Fig 7).

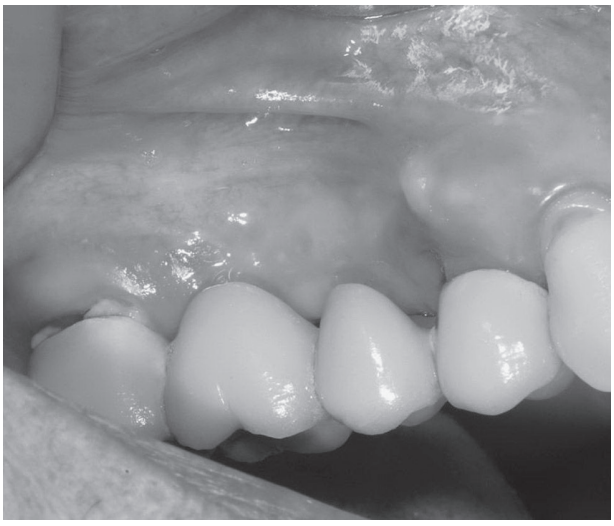

Fig 6 Intraoral view 2 years postoperatively. There is no sign of recurrence.

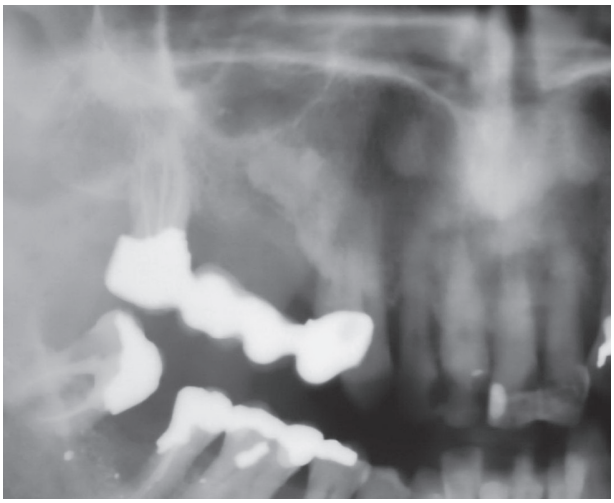

Fig 7 Panoramic radiograph 2 years postoperatively. Notice normal bone healing.

DISCUSSION

Peripheral osteoma is more common in the sinuses, especially the frontal, followed by the ethmoidal and maxillary. Rare sites are the osseous portion of the external auditory canal, orbit, temporal bone, and pterygoid processes.[3,5] Peripheral osteomas of the jaws are uncommon with the mandible usually affected.

Solitary peripheral osteomas of the maxilla are very rare lesions.[2,5,8,11] Johann et al[5] found six well-documented cases of maxillary peripheral osteomas in a review of the English literature during the last 76 years, excluding cases in the maxillary sinuses as well as osteomas related to Gardner's syndrome. Four cases involved the alveolar process and two cases the hard palate. The female to male ratio was

Table 1. Data From the Literature Concerning Six Cases of Peripheral Osteoma of the Maxillary Alveolar Process

Case No.	*Age/ (Years)*	*Sex*	*Size (cm)*	*Presenting Symptom*	*Author(s)*
1	50	F	—	—	Seward (1965)[11]
2–4	—	2 M,1 F	—	—	Sayan et al (2002)[3]
5	76	F	1.0	Local sensitivity	Woldenberg et al (2005)[12]
6	16	F	2.5 ×1.5	Deformity	Dalambiras et al (2005)[2]

2:1, and the only patient in whom age was cited was 50 years old. Woldenberg et al[12] studied 14 consecutive cases of peripheral osteomas of the maxillofacial region. One case of a solitary peripheral osteoma in the buccal aspect of the maxilla in a 76-year-old woman was recorded. Dalambiras et al[2] reported a case of solitary peripheral osteoma of the anterior right maxilla associated with an impacted permanent maxillary canine in a 16-year-old girl. From the literature review, it seems that only six cases of solitary peripheral osteomas of the maxillary alveolar process have been reported up to 2006 (Table 1).

Pathogenesis of the peripheral osteoma is controversial. Peripheral osteomas of the sinuses develop in association with inflammatory sinus polyps, whereas peripheral osteomas of the osseous portion of the external auditory canal have been reported in cold water swimmers and divers and have been experimentally induced in animals using cold water rinses.[13,14] Thus, most cases of peripheral osteomas of the jaws are probably reactive, inflammatory hyperplasias induced by a combination of trauma and muscle traction.[6] In fact, most cases of jaw lesions are located in the lower border or buccal aspect of the mandible, sites more susceptible to trauma than the lingual aspect of the mandible or the buccal aspect of the maxilla. Furthermore, most cases are not randomly distributed, but occur in close proximity to areas of muscle attachment. It is suggested that a minor trauma, unlikely to be remembered by a patient, may cause subperiosteal edema or hemorrhage and muscle traction that could locally elevate the periosteum. These two elements might initiate an osteogenic reaction that could be preserved by continuous muscle traction.[3]

The developmental theory of pathogenesis is not supported by the common development of peripheral osteomas in adults and not children, whereas the limited growth potential of the lesions as well as infrequent recurrence are against a true neoplastic origin.[2,12,15]

Differential diagnosis of a solitary peripheral osteoma may include focal sclerosing osteomyelitis, peripheral ossifying fibroma, exostoses, sessile osteochondroma, periosteal osteoblastoma, odontoma, and malignant tumors such as parosteal osteosarcoma.[2,5,16,17] Nevertheless, the most similar entities that may occur in the buccal plate of the alveolar ridge in the bicuspid area of the maxilla are exostoses. However, exostoses are described as multiple, rounded, nodular bony outgrowths over the apices of the teeth and below the mucobuccal fold.[18,19] In our case, the lesion appeared as a single, intraosseous, sessile, sclerotic, protuberant growth expanding toward the alveolar ridge. Moreover, exostoses appear in radiographs as well-defined radiopacities, whereas in our case, the lesion appeared as an ill-defined radiopaque mass.

Multiple osteomas are a sign of Gardner's syndrome, a phenotypic variant of familial adenomatous polyposis coli. It is inherited with the autosomic-dominant type and characterized by germline mutations in the 5q chromosomal area of the adenomatous polyposis coli gene. Other signs include adenomatous intestinal polyps that possess a great potential of malignant transformation, cutaneous epidermal cysts, odontomas, supernumerary and impacted teeth. Osteomas of the maxillofacial region are an early manifestation of Gardner's syndrome; thus, dentists and oral surgeons should address patients for further medical investigation, because early diagnosis is fundamental for prognosis.[20–22]

Peripheral osteomas are usually surgically excised for cosmetic or diagnostic purposes, when the lesion is symptomatic or actively growing, and when function is compromised.[11] Recurrence is very rare and there are no reports of malignant transformation.[2,12,15]

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The Importance of the Head and Neck Region in Regression of Advanced MCC: A Clinical Report

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Abstract: This paper presents the case of a 76-year-old woman who experienced a total regression of a Merkel cell carcinoma (MCC). The primary site of the tumor was on her right eyebrow. After this lesion was excised, the patient presented a massive locoregional metastasis on the right parotid gland and the laterocervical lymph nodes. No distant metastases were detected. An incisional biopsy into the right parotid gland confirmed the diagnosis of MCC metastasis. No surgical treatment was prescribed because of the advanced stage of the disease. Spontaneous total regression on the parotid and the neck mass was observed within 3 months. This is the 15th case of spontaneous regression in total and the 14th case with a site of origin in the head and neck region.

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Key Words: Merkel cell carcinoma, spontaneous regression, biopsy

Merkel cell carcinoma (MCC) is a rare primary neuroendocrine carcinoma of the skin. The clinical behavior is characterized by high incidence of local recurrence (27–60%), of lymph node metastases (45–91%), and of distant metastases in the liver, bone, brain, lung, or skin (18–52%).[1] The incidence of disease-related death ranges from 35% to almost 50%.[2–4] Despite its highly malignant nature, 14 cases of spontaneous regression have been reported to date. Only 12 can be classified as complete spontaneous regression after the performance of only a biopsy.[2,3,5,6,8–13] We describe the clinical course of patient with advanced stage II MCC of the head and neck district that showed complete spontaneous regression after biopsy, and we emphasize the correlation between the head and neck area and the regression (Figs 1–4).

Clinical Report

A 76-year-old white woman came to the Department of Dermatology at the University of Rome, ''La Sapienza'' in October of 2005. She had a painless, pink-red, dome-shaped nodule on her right eyebrow that had first appeared 3 months earlier and was growing rapidly. The greatest diameters of the nodule ranged from 0.5 to 0.8 cm. There were no palpable adenopathies.

The lesion was excised under local anesthesia on October 19, 2005. The histopathologic examination showed that the neoplasm had the characteristic pattern of MCC.

On 29 November of 2005, total body and neck CTs with contrast were performed. They revealed no local relapse or metastases.

Four months later, in March of 2006, a nodular lesion appeared in front of her right ear. Scintigraphy with the radiolabeled somatostatin analogue octreotide showed a suspicious accumulation of radioactivity in the mastoid area. On March 24, a computed tomography (CT) scan of the head and neck and total body demonstrated a solid tumor in place of the right parotid gland. The greatest diameters of the tumor ranged from 50 to 30 mm with positive and inhomogeneous enhancement. Retromandibular lymph nodal metastases were also present and included the first to fifth level of the neck.[14] There were, however, no distant metastases (Figs 1 and 2).

Surgery was excluded because of the advanced stage of the disease. Before chemotherapy, the