Metastatic and salivary-type carcinomas. *Head Neck*. 2013;35:895-901.
9. Li Y, Li LJ, Huang J, Han B, Pan J. Central malignant salivary gland tumors of the jaw: retrospective clinical analysis of 22 cases. *J Oral Maxillofac Surg*. 2008;66:2247-2253.
10. Han J, Gu T, Yang X, et al. Primary intraosseous adenoid cystic carcinoma of the mandible: a comprehensive review and analysis of four new cases with emphasis on morphologic, immunophenotypic, and molecular characteristics. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2017;123:365-373.
11. Bouaichi A, Aimad-Eddine S, Mommers XA, Ella B, Zwetyenga N. Intra-mandibular adenoid cystic carcinoma. *Rev Stomatol Chir Maxillo-Fac Chir Orale*. 2014;115:100-104.
12. Stenman G, Licitra L, Said-Al-Naief N, van Zante A, Yarbrough W. WHO classification of head and neck tumours. In: El-Naggar AK, Chan JKC, Grandis J, Takata T, Slootweg PJ, eds. *World Health Organization Classification of Tumours*. ed 4. Lyon, France: International Agency for Research on Cancer; 2017:164-165.
13. Hellquist H, Skalova A. Adenoid cystic carcinoma. In: Skalova A, ed. *Histopathology of the Salivary Glands*. Berlin, Heidelberg: Springer Berlin Heidelberg; 2014:221-253.
14. Simpson RH, Skalova A, Di Palma S, Leivo I. Recent advances in the diagnostic pathology of salivary carcinomas. *Virchows Arch*. 2014;465:371-384.
15. Argyris PP, Wetzel SL, Greipp P, et al. Clinical utility of myb rearrangement detection and p63/p40 immunophenotyping in the diagnosis of adenoid cystic carcinoma of minor salivary glands: a pilot study. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2016;121:282-289.
16. Rooper L, Sharma R, Bishop JA. Polymorphous low grade adenocarcinoma has a consistent p63+/p40- immunophenotype that helps distinguish it from adenoid cystic carcinoma and cellular pleomorphic adenoma. *Head Neck Pathol*. 2015;9:79-84.
17. Rettig EM, Tan M, Ling S, et al. MYB rearrangement and clinicopathologic characteristics in head and neck adenoid cystic carcinoma. *Laryngoscope*. 2015;125:E292-E299.

CLINICAL PATHOLOGIC CONFERENCE CASE 4: RECURRENT GINGIVAL GROWTH IN THE ANTERIOR MAXILLA

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Clinical Presentation: A 57-year-old female presented to an oral surgeon with recurrence of an asymptomatic, slowly enlarging, firm swelling involving the anterior maxillary gingiva. Six excisional biopsies had previously been performed by several different clinicians over the preceding decade. The histologic diagnoses included multiple diagnoses of fibrous epulis and peripheral ossifying fibroma. The patient had no contributory medical history. Extraoral examination was unremarkable. Intraoral examination showed a 2.0 × 1.5 × 1.5 cm sessile nodular mass involving the buccal and palatal attached gingiva and extending from the extraction site of right maxillary central incisor to the interdental papilla between the left maxillary central and lateral incisor (Figure 1). The affected area showed normal mucosal coloration with a stippled surface texture. There was no apparent ulceration, hemorrhage, or exudate associated with the lesion. The left maxillary lateral incisor was noted to be displaced coronally and buccally compared with the contralateral lateral incisor. All remaining maxillary teeth were confirmed to be vital. Periapical radiographs showed generalized alveolar bone loss. In the area of the lesion, the alveolar crest mesial to the left maxillary central incisor approximated the level of the central incisor's cementoenamel junction, which was considerably higher than the overall alveolar crest height of the adjacent maxillary teeth. At the superior aspect of the bony crest, faint spicules were identified. A diagonal, linear radiolucency was present just inferior to the crest of bone (Figure 2). There was no obvious cortical erosion or thickening of the soft tissue shadow in the area of the lesion. No widening of the periodontal ligament (PDL) spaces or loss of lamina dura was identified. There was no displacement or resorption of tooth roots; however, the left maxillary lateral incisor appeared to be displaced coronally. Several teeth showed evidence of previous endodontic treatment and restoration with porcelain crowns.

Differential Diagnosis: The clinical presentation of a sessile, smooth-surfaced, normocolored nodule involving the gingiva suggested that the lesion was composed of tissue(s) located below the epithelial surface. Therefore, the differential diagnosis included reactive and neoplastic proliferations of mesenchymal tissues typically located below the surface epithelium in the gingiva. The differential diagnosis list included common gingival reactive connective tissue lesions: peripheral fibroma, pyogenic granuloma, peripheral ossifying fibroma (POF), and peripheral giant cell granuloma (PGCG), as well as uncommon benign mesenchymal neoplasms or tumor-like conditions, such as neurofibroma, schwannoma, and vascular malformation. Moreover, given the location, a peripheral odontogenic tumor was also included in the differential diagnosis list.

Fibroma is the most common reactive proliferation or "tumor" of the oral cavity and represents a focal hyperplasia of fibrous connective tissue in response to chronic irritation or trauma.[1,2] This lesion most commonly occurs at the level of the occlusal plane in the buccal mucosa; however, the gingiva is also a possible location. A typical fibroma presents as a well-defined, pink, smooth-surfaced, sessile nodule of fibrous consistenty.[3] Pyogenic granuloma represents an exuberant, hypervascular proliferation of granulation tissue in response to low-grade irritation or trauma.[4] Clinically, it appears as a rapidly growing smooth or lobulated erythematous mass that is usually painless, bleeds easily, and most commonly involves the gingiva.[2] PGCG is another common reactive lesion of the oral cavity that occurs exclusively in the gingiva or the edentulous alveolar ridge. This lesion usually appears as a red-blue or purple nodule that might be sessile or pedunculated. The average age at onset for PGCG is 46 years, and a recurrence rate of 17.5% has been reported.[5] Finally, POF completes the list of the 4 most common reactive localized overgrowths of the gingiva. POF is believed to originate from the periodontal ligament; consequently, it occurs exclusively on the gingiva. It shows a predilection for females in the second decade of life and tends to occur in the maxilla, particularly the incisor−cuspid region.[6] The POF usually appears as a sessile or pedunculated nodule that varies in color from pink to red and usually exhibits an ulcerated surface.[7] Although these lesions are usually less than 2 cm in diameter, rare "giant" POFs measuring greater than 2.5 cm and causing displacement of teeth have been described.[8] Furthermore, recurrences have been reported for POFs, and the recurrence rate has been estimated to be around

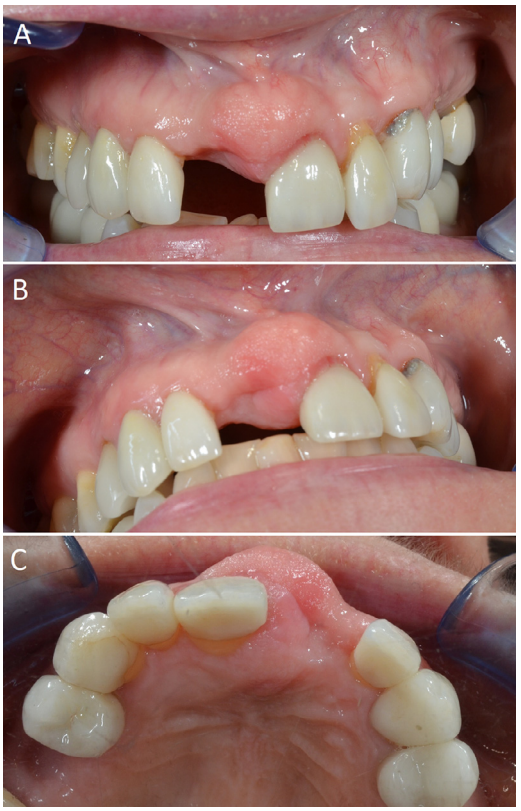

Fig. 1. **(A)** Intraoral examination showed a normocolored sessile mass involving the buccal attached gingiva extending from the extraction site of tooth #8 to the interdental papilla between teeth #9 and #10. **(B)** There were focal areas of mild erythema involving the extraction site of tooth #8 and the buccal gingival margin of tooth #9. **(C)** The mass also extended to the mesial aspect of tooth #9 and the palatal gingiva.

8% to 16%. The recurrence may be caused by incomplete removal of the lesion or failure to eliminate potential local irritants. Furthermore, it has been generally recommended that the surgical excision of a POF should include underlying periosteum and affected PDL to decrease the likelihood of a recurrence.[6,7] Although these 4 reactive proliferations are very common on the gingiva, the fact that the lesion in the present case appeared normal in color and not conspicuously inflamed made a pyogenic granuloma or a PGCG an unlikely diagnosis. A fibroma was also considered unlikely, given the numerous recurrences of the lesion. In contrast, occasional POFs have been reported to recur multiple times. Furthermore, it is plausible that given the location of this particular lesion, the previous excisions had been too conservative in an attempt to avoid creating a gingival defect and, consequently, failed to completely remove the underlying reactive PDL.

Because the lesion was overlying the area of the incisive canal and clinically appeared to be mesenchymal in nature, benign peripheral nerve sheath tumors, such as a neurofibroma or schwannoma, and a vascular malformation (VM) were also considered. Oral neural lesions are rare, representing only 0.2% to 0.6% of the total cases received by oral pathology biopsy services.[9-12] Oral neural lesions are more commonly seen in females and tend to affect patients age 43.3 years on average.[12] The tongue, palate, gingiva, and lips are among the most commonly involved oral sites.[9,11,12] In fact, 2 case series found that gingiva and the alveolar mucosa were the most frequent intraoral sites affected by neurofibromas.[12,13] Neurofibroma is the most common type of intraoral peripheral nerve sheath tumor and may occur as a solitary lesion or as multiple lesions associated with neurofibromatosis type I.[10,12,13] It is an unencapsulated tumor consisting of a mixture of Schwann cells, perineurial cells, and fibroblasts.[14] Oral neurofibromas typically appear as a slow-growing, painless, pale pink, and nonulcerated nodules or masses.[12,15-17] Schwannomas or neurilemmomas are benign neural neoplasms arising from Schwann cells. Although these tumors are rare, one-fourth to one-half of the cases occur in the head and neck region.[12] The lips and the tongue are the sites of

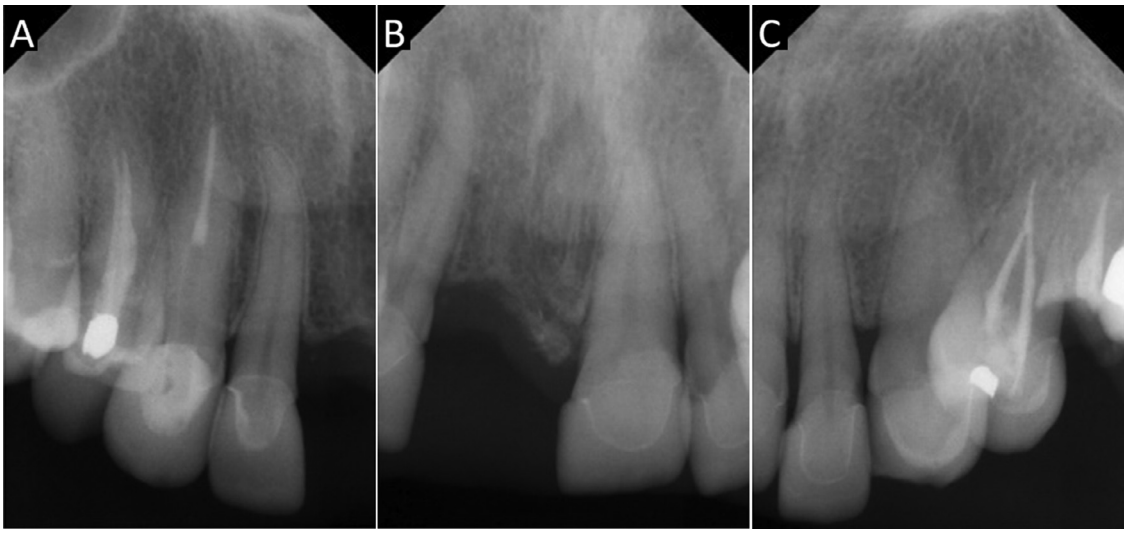

Fig. 2. Periapical radiographs showing generalized alveolar bone loss. In the area of the lesion, the alveolar crest appeared higher compared with the alveolar crest level of adjacent maxillary teeth.

predilection for this neural tumor, which typically presents as a well-defined, painless, and slow-growing mass.[12,18] Both neurofibroma and schwannoma are treated with surgical excision, and recurrence is rare.[13,18]

VMs are present at birth and are slow growing, infiltrative, and even destructive. In contrast to hemangiomas of infancy, which grow rapidly in the first 6 months of life and then start to involute over several years, VMs do not regress and continue to expand with time.[19,20] In the current case, the patient was 57 years of age. If the lesion was indeed vascular in nature, it would more likely represent a VM rather than a true hemangioma. VMs are irregular vascular networks, which are classified according to their particular blood vessel type. Clinically, depending on the depth of the abnormal vasculature, VMs may appear normal in color or exhibit a bluish or deep-purple discoloration. The lesions fill with dependency and are compressible. VMs are usually ill-defined lesions, and their true extent is difficult to determine intraoperatively because of bleeding; hence, their treatment is usually a challenge and often requires a multidisciplinary approach and more than one treatment modality, which may include surgery,Neodymium-doped Yttrium Aluminum Garnet laser therapy, and/or sclerotherapy.[20,21]

Peripheral odontogenic tumors are rare gingival lesions, representing 0.5% of specimens received by oral pathology biopsy services and 1.5% of all gingival lesions.[22-24] Of all odontogenic tumors, odontogenic fibroma, ameloblastoma, and calcifying odontogenic cyst are the most common to occur in the gingival soft tissues.[22,24] Clinically, these tumors occur in patients in the fourth through seventh decades of life, exhibit a predilection for the incisor/canine and premolar regions of the mandible, and typically mimic common gingival reactive lesions, such as a fibroma or pyogenic granuloma.[22,24] Peripheral odontogenic tumors are usually treated appropriately with local surgical excision, and these tumors in general exhibit an innocuous biologic behavior, even in the case of a peripheral ameloblastoma.[25,26] Indeed, peripheral ameloblastomas show a much lower recurrence rate compared with intraosseous ameloblastomas.[27] Although it is widely accepted that these tumors also exhibit benign clinical behavior, a few studies have demonstrated a significant recurrence rate for peripheral odontogenic fibromas, which, in one large case series study, was as high as 50%.[28,29]

Given the fact that the current lesion had recurred several times, a low-grade malignancy that was initially misdiagnosed was also entertained as a diagnostic possibility. Although both the clinical and radiographic features of the lesion were not overtly ominous indications of a malignancy, the higher level of the alveolar crest in the mesial aspect of tooth #9 compared with the alveolar crest level of all the surrounding maxillary teeth raised the concern that the lesion could represent a bone-forming tumor, particularly parosteal osteosarcoma. Parosteal osteosarcoma is a low-grade, surface (juxtacortical) variant of osteosarcoma, which is extremely rare in the jaws.[30] This specific type of osteosarcoma shows a slight male predilection, and there is nearly equal distribution between the maxilla and the mandible. It appears radiographically as a well-defined opaque mass that is attached to the cortical bone by a short pedicle.[31] The clinical presentation is usually of a painless, firm swelling, which, on occasion, may become ulcerated.[32] Histopathologically, it typically appears as irregular bone trabeculae intermixed with a spindle cell stroma showing minimal atypia and rare mitotic figures. Because of the bland histopathologic, clinical, and radiographic appearances, it is quite common for these lesions to be misdiagnosed initially as benign lesions.[31] Parosteal osteosarcoma of the jaws seems to behave in a manner similar to its long bone counterpart, growing slowly and showing minimal potential for metastasis; however, it does tend to recur after a simple local excision.[32]

Diagnosis: Incisional biopsy was completed by the oral surgeon and the sample submitted for histologic examination. The 1.7 × 1.4 × 1.3 cm portion of soft tissue was processed routinely and stained with hematoxylin and eosin. At low power, the specimen showed unremarkable gingival mucosa overlying fibrous connective tissue proliferation containing fragments of bone that involved the deep biopsy margin (Figure 3A). Higher power showed fragments of woven bone with minimal osteoblastic rimming, surrounded by a moderately cellular fibrous spindle cell

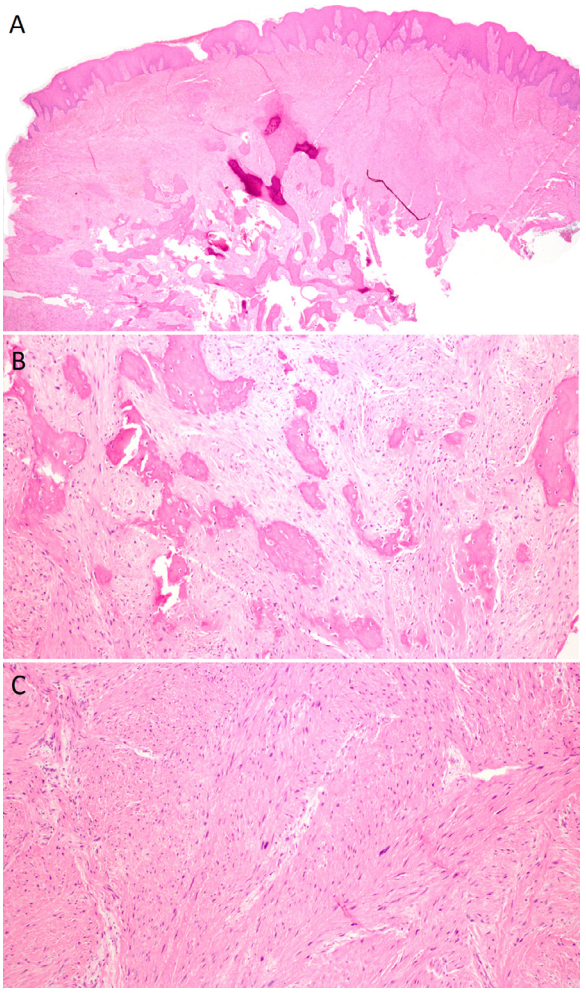

Fig. 3. Incisional biopsy. **(A)** Nodular fragment of mucosa showing a fibrous connective tissue proliferation containing irregular trabeculae of bone involving the deep biopsy margin (original magnification × 40). **(B)** Small trabeculae of woven bone with minimal osteoblastic rimming surrounded by a moderately cellular fibrous stroma (original magnification × 200). **(C)** Focally, the fibrous spindle cell stroma became more cellular and assumed a fascicular growth pattern. Occasional atypical, enlarged and hyperchromatic nuclei were identified (original magnification × 200).

stroma with minimal vascularity and no extravasated blood or giant cells (Figure 3B). No odontogenic epithelium was identified, and there was minimal osteoid production. Focally, the fibrous spindle cell stroma produced a storiform to fascicular growth pattern with rare cells showing enlarged, pleomorphic, and hyperchromatic nuclei (Figure 3C). Mitotic figures were inconspicuous. Mouse double minute 2 homolog (MDM2; Figure 4A) and special AT-rich sequence-binding protein 2 (SATB2; Figure 4B) immunohistochemical stains were completed and both showed diffuse nuclear positivity. Fluorescence in situ hybridization confirmed the *MDM2* gene amplification and led to a diagnosis of low-grade osteosarcoma, parosteal subtype.

Management: Surgical workup, including helical computed tomography and magnetic resonance imaging, could not detect the small tumor focus. No metastases were identified. The patient underwent a partial maxillectomy of the anterior premaxilla, with radial forearm flap reconstruction. Microscopic examination of the resection specimen confirmed destructive and infiltrative spindle cell proliferation involving the alveolar bone of the premaxilla consistent with low-grade osteosarcoma, parosteal subtype (Figure 5A). Foci of osteoid production (Figure 5B) and increased cellularity (Figure 5C) were identified throughout the specimen. Mitotic figures remained inconspicuous, and necrosis was not identified. After 6 months of follow-up, the patient remained disease free, with excellent healing of the surgical site and no evidence of local recurrence or distant metastases.

Discussion: Osteosarcoma, a malignant neoplasm of bone, is rare in the gnathic bones, constituting 5% to 13% of skeletal osteosarcomas[33-35] and 1% of head and neck malignancies.[34,36] The majority of osteosarcomas involving the gnathic bones are high-grade types, with low-grade variants occurring very rarely.[35] Of the low-grade, well-differentiated variants reported in the head and neck, intramedullary osteosarcoma occurs more frequently compared with parosteal (juxtacortical) osteosarcoma.[35] Low-grade osteosarcoma of the gnathic bones occurs over a broad age range with most reported cases occurring in the third and fourth decades.[37,38] The mandible is affected more frequently compared with the maxilla, and most present with longstanding, asymptomatic swellings of the affected jaw.[37,38] Because of their slow growth, benign radiographic presentation, and well-differentiated histologic appearance, these malignancies are often mistaken for reactive proliferations or benign neoplasms, such as peripheral ossifying fibroma, osteomas, and benign fibro-

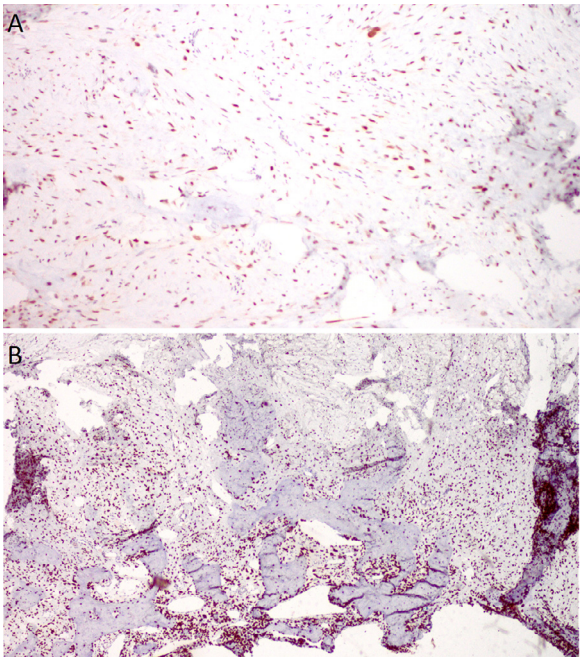

Fig. 4. **(A)** Immunohistochemical staining with MDM2 showed diffuse nuclear positivity (original magnification × 200). **(B)** Immunohistochemical staining with SATB2 showed strong diffuse nuclear positivity (original magnification × 40).

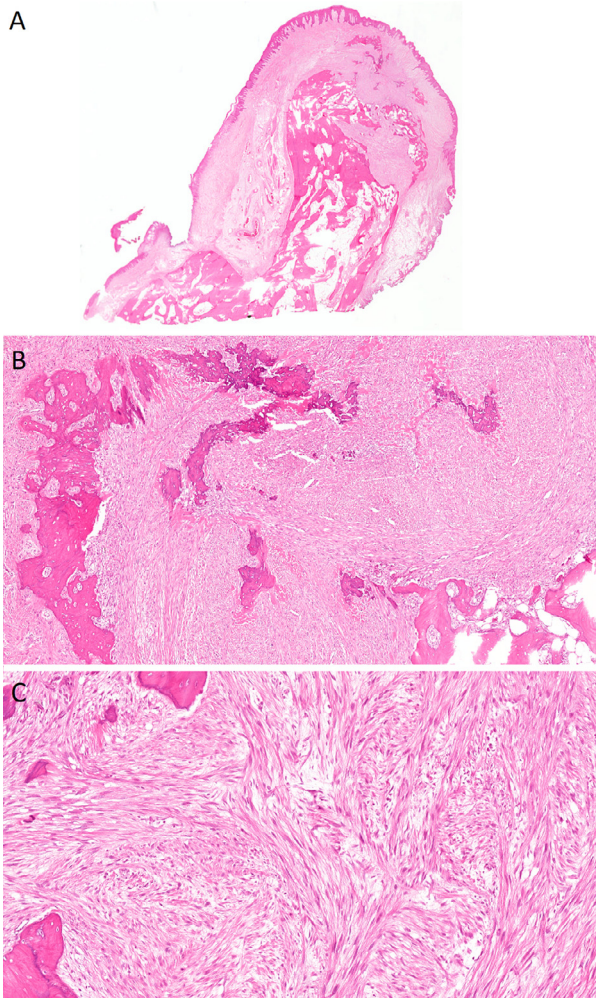

Fig. 5. Resection specimen. **(A)** Destructive and infiltrative spindle cell proliferation forming well-differentiated bone, which is seen attached to the underlying alveolar bone cortex by a stalk, consistent with surface low-grade osteosarcoma, parosteal subtype (original magnification × 20). **(B)** Areas of irregular osteoid were seen throughout the lesion (original magnification × 100). **(C)** Significantly cellular areas showing spindle-shaped cells with occasional enlarged, atypical nuclei were seen (original magnification × 400).

osseous lesions of the jaw.[32,34,35,39] MDM2 and CDK4 immunohistochemical stains are useful in the differentiation of low-grade osteosarcoma from benign lesions of the gnathic bones.[40,41] Together, these stains show 100% sensitivity and 97.5% specificity for low-grade osteosarcoma of the gnathic bones.[41] In select cases, SATB2 can also be useful in confirming the osteoblastic lineage of the well-differentiated malignant cell population.[42-44] Wide surgical excision with negative margins is the treatment of choice,[35] resulting in a 5-year survival rate of 80%.[37] Positive margins after excision have been associated with lesion recurrence.[35] Currently, there is no evidence to support the routine use of radiation therapy and/or chemotherapy in the treatment of low-grade osteosarcoma of the gnathic bones.[35,37]

Although diagnosis based on the clinical, radiographic, and histologic features of low-grade osteosarcoma of the gnathic bones can be very challenging, attention to subtle changes are vital to the identification of this lesion. Multiple clinical recurrences of the same lesion are always a sign that the differential diagnosis and treatment approach should be re-evaluated. Interdental alveolar bone height that appears focally higher compared with adjacent sites should alert the clinician to the possibility of a lesion that may be causing deposition of calcified material. Histologically, all lesions associated with spindle cell proliferations should be thoroughly assessed for aggressive growth patterns, areas of increased cellularity and any cellular or nuclear pleomorphism.

References

1. Amirchaghmaghi M, Mohtasham N, Mosannen Mozafari P, Dalirsani Z. Survey of reactive hyperplastic lesions of the oral cavity in Mashhad, Northeast Iran. *J Dent Res Dent Clin Dent Prospects*. 2011;5:128-131.
2. Gonsalves WC, Chi AC, Neville BW. Common oral lesions: Part II. Masses and neoplasia. *Am Fam Physician.* 2007;75:509-512.
3. Holmstrup P, Plemons J, Meyle J. Non-plaque-induced gingival diseases. *J Clin Periodontol.* 2018;45:S28-S43.
4. Gordon-Nunez MA, de Vasconcelos Carvalho M, Benevenuto TG, Lopes MF, Silva LM, Galvao HC. Oral pyogenic granuloma: a retrospective analysis of 293 cases in a Brazilian population. *J Oral Maxillofac Surg.* 2010;68:2185-2188.
5. Lester SR, Cordell KG, Rosebush MS, Palaiologou AA, Maney P. Peripheral giant cell granulomas: a series of 279 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2014;118:475-482.
6. Buchner A, Hansen LS. The histomorphologic spectrum of peripheral ossifying fibroma. *Oral Surg Oral Med Oral Pathol.* 1987;63:452-461.
7. Cuisia ZE, Brannon RB. Peripheral ossifying fibroma—a clinical evaluation of 134 pediatric cases. *Pediatr Dent.* 2001;23:245-248.
8. Childers EL, Morton I, Fryer CE, Shokrani B. Giant peripheral ossifying fibroma: a case report and clinicopathologic review of 10 cases from the literature. *Head Neck Pathol.* 2013;7:356-360.
9. Franco T, de Freitas Filho SA, Muniz LB, de Faria PR, Loyola AM, Cardoso SV. Oral peripheral nerve sheath tumors: a clinicopathological and immunohistochemical study of 32 cases in a Brazilian population. *J Clin Exp Dent.* 2017;9: e1459-e1465.
10. Alotaibi O, Al Sheddi M. Neurogenic tumors and tumor-like lesions of the oral and maxillofacial region: a clinicopathological study. *Saudi Dent J.* 2016;28:76-79.
11. Salla JT, Johann AC, Garcia BG, Aguiar MC, Mesquita RA. Retrospective analysis of oral peripheral nerve sheath tumors in Brazilians. *Braz Oral Res.* 2009;23:43-48.
12. Alotaiby FM, Fitzpatrick S, Upadhyaya J, Islam MN, Cohen D, Bhattacharyya I. Demographic, clinical and histopathological features of oral neural neoplasms: a retrospective study. *Head Neck Pathol.* 2018 Jun 21. doi: 10.1007/s12105-018-0943-1. [Epub ahead of print].
13. Marocchio LS, Oliveira DT, Pereira MC, Soares CT, Fleury RN. Sporadic and multiple neurofibromas in the head and neck region: a retrospective study of 33 years. *Clin Oral Investig.* 2007;11:165-169.
14. Chrysomali E, Papanicolaou SI, Dekker NP, Regezi JA. Benign neural tumors of the oral cavity: a comparative immunohistochemical study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1997;84:381-390.
15. Ohno J, Iwahashi T, Ozasa R, Okamura K, Taniguchi K. Solitary neurofibroma of the gingiva with prominent differentiation of Meissner bodies: a case report. *Diagn Pathol.* 2010;5:61.
16. Mahmud SA, Shah N, Chattaraj M, Gayen S. Solitary encapsulated neurofibroma not associated with neurofibromatosis-1 affecting tongue in a 73-year-old female. *Case Rep Dent.* 2016;2016:3630153.
17. Garcia de Marcos JA, Dean Ferrer A, Alamillos Granados F, et al. Gingival neurofibroma in a neurofibromatosis type 1 patient. *Med Oral Patol Oral Cir Bucal.* 2007;12:E287-E291.
18. Hsu YC, Hwang CF, Hsu RF, Kuo FY, Chien CY. Schwannoma (neurilemmoma) of the tongue. *Acta Otolaryngol.* 2006;126:861-865.
19. Adams DM, Lucky AW. Cervicofacial vascular anomalies. I. Hemangiomas and other benign vascular tumors. *Semin Pediatr Surg.* 2006;15:124-132.
20. Richter GT, Friedman AB. Hemangiomas and vascular malformations: current theory and management. *Int J Pediatr.* 2012;2012:645678.
21. Buckmiller LM, Richter GT, Suen JY. Diagnosis and management of hemangiomas and vascular malformations of the head and neck. *Oral Dis.* 2010;16:405-418.
22. Buchner A, Merrell PW, Carpenter WM. Relative frequency of peripheral odontogenic tumors: a study of 45 new cases and comparison with studies from the literature. *J Oral Pathol Med.* 2006;35:385-391.
23. Ide F, Obara K, Mishima K, et al. Peripheral odontogenic tumor: a clinicopathologic study of 30 cases. General features and hamartomatous lesions. *J Oral Pathol Med.* 2005;34:552-557.
24. Manor Y, Mardinger O, Katz J, Taicher S, Hirshberg A. Peripheral odontogenic tumours—differential diagnosis in gingival lesions. *Int J Oral Maxillofac Surg.* 2004;33:268-273.
25. Buchner A, Sciubba JJ. Peripheral epithelial odontogenic tumors: a review. *Oral Surg Oral Med Oral Pathol.* 1987;63:688-697.
26. Philipsen HP, Reichart PA, Nikai H, Takata T, Kudo Y. Peripheral ameloblastoma: biological profile based on 160 cases from the literature. *Oral Oncol.* 2001;37:17-27.
27. Nauta JM, Panders AK, Schoots CJ, Vermey A, Roodenburg JL. Peripheral ameloblastoma. A case report and review of

the literature. *Int J Oral Maxillofac Surg*. 1992;21:40-44.
28. Ritwik P, Brannon RB. Peripheral odontogenic fibroma: a clinicopathologic study of 151 cases and review of the literature with special emphasis on recurrence. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2010;110:357-363.
29. Daley TD, Wysocki GP. Peripheral odontogenic fibroma. *Oral Surg Oral Med Oral Pathol.* 1994;78:329-336.
30. Puranik SR, Puranik RS, Ramdurg PK, Choudhary GR. Parosteal osteosarcoma: report of a rare juxtacortical variant of osteosarcoma affecting the maxilla. *J Oral Maxillofac Pathol.* 2014;18:432-436.
31. Huang TC, Monsour PA, Chahoud CD. Parosteal osteosarcoma: report of a case and review of the literature. *Aust Dent J.* 2010;55:86-91.
32. Hewitt KM, Ellis G, Wiggins R, Bentz BG. Parosteal osteosarcoma: case report and review of the literature. *Head Neck*. 2008;30:122-126.
33. Mardinger O, Givol N, Talmi YP, Taicher S. Osteosarcoma of the jaw. The Chaim Sheba Medical Center experience. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2001;91:445-451.
34. Sinha R, Roy Chowdhury SK, Chattopadhyay PK, Rajkumar K. Low-grade osteosarcoma of the mandible. *J Maxillofac Oral Surg*. 2010;9:186-190.
35. Demicco EG, Deshpande V, Nielsen GP, Kattapuram SV, Rosenberg AE. Well-differentiated osteosarcoma of the jaw bones: a clinicopathologic study of 15 cases. *Am J Surg Pathol.* 2010;34:1647-1655.
36. Thiele OC, Freier K, Bacon C, Egerer G, Hofele CM. Interdisciplinary combined treatment of craniofacial osteosarcoma with neoadjuvant and adjuvant chemotherapy and excision of the tumour: a retrospective study. *Br J Oral Maxillofac Surg*. 2008;46:533-536.
37. Diniz AF, Filho JA, Alencar Rde C, et al. Low-grade central osteosarcoma of the mandible: a case study report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2007;103:246-252.
38. Bertoni F, Bacchini P, Fabbri N, et al. Osteosarcoma. Low-grade intraosseous-type osteosarcoma, histologically resembling parosteal osteosarcoma, fibrous dysplasia, and desmoplastic fibroma. *Cancer*. 1993;71:338-345.
39. Bhowmik B, Naikmasur VG, Acharya S, Tayaar SA, Bangera R, Dinesh US. Low grade osteosarcoma of maxilla: report of a case and review of literature. *J Oral Maxillofac Surg Med Pathol.* 2014;26:590-595.
40. Dujardin F, Binh MB, Bouvier C, et al. MDM2 and CDK4 immunohistochemistry is a valuable tool in the differential diagnosis of low-grade osteosarcomas and other primary fibro-osseous lesions of the bone. *Mod Pathol.* 2011;24:624-637.
41. Yoshida A, Ushiku T, Motoi T, et al. Immunohistochemical analysis of MDM2 and CDK4 distinguishes low-grade osteosarcoma from benign mimics. *Mod Pathol.* 2010;23:1279-1288.
42. Chen CY, Zhang HZ, Jiang ZM, Zhou J, Chen J, Liu L. Value of MDM2, CDK4 and SATB2 immunohistochemistry in histologic diagnosis of low-grade osteosarcoma. *Zhonghua Bing Li Xue Za Zhi.* 2016;45:387-392 [in Chinese].
43. Conner JR, Hornick JL. SATB2 is a novel marker of osteoblastic differentiation in bone and soft tissue tumours. *Histopathology*. 2013;63:36-49.
44. Falconieri G, Cataldi P, Kavalar R, Stitic V, Luzar B. Cutaneous osteoblastic osteosarcoma: report of 2 new cases integrated with SATB2 immunohistochemistry and review of the literature. *Am J Dermatopathol*. 2016;38:824-831.

CLINICAL PATHOLOGY CONFERENCE CASE 5: BILATERAL MAXILLARY SINUS RESORPTIVE DISEASE WITH PALATAL ULCERATION IN A 42-YEAR-OLD PATIENT

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Clinical Presentation: A 42-year-old female presented with bilateral periodontal swellings of 6 months' duration in the maxillary area palatal to the maxillary molar and premolar teeth. The lesions had been treated by a general dentist as localized periodontal disease, and because of local pain, root canal treatment of tooth #2 was administered. As a result of medial expansion of the lesions toward the hard palate (Figure 1) over the next few months and the occurrence of pus-like discharge, orthopantomography (Figure 2) and computed tomography (CT) (Figure 3) were performed, and the examinations showed bilateral lytic lesions of the hard palate involving the roots of teeth # 4 to #14, respectively. The patient was then referred to the Odontostomatology Clinic of the University of Bari, where clinical inspection confirmed the presence of bilateral and slightly elevated, erosive lesions of the hard palate. In view of the radiologic features, total body CT was performed, and no additional lesions were identified. The remaining clinical history and laboratory tests were noncontributory, with values within normal limits.

Differential Diagnosis: On the basis of the presenting signs and symptoms, review of the medical history, and analysis of the available information, including the clinical and radiographic features, several entities were considered in the differential diagnosis of the current case.

The presence of an odontogenic infection with subsequent maxillary sinus extension, also referred to as *odontogenic sinusitis*, has been attributed to approximately 10% to 12% and up to 68% of all sinusitis cases.[1,2] It has variable clinical and radiographic features, ranging from minimal signs and symptoms with focal inflammation and radiograph evidence of sinus lining thickening (which can be further confirmed with advanced CT) to severe, full-blown, and progressive rhinosinusitis, with potential progression to orbital cellulitis, blindness, meningitis, and cavernous sinus fibrosis, among other complications.[3] The majority of cases of odontogenic sinusitis are triggered iatrogenically, secondary to injury of the mucoperiosteum and violation of the Schneiderian membrane lining of the maxillary sinus, or occur after dental procedures, such as extractions, implant placement, sinus augmentation surgery, and orthognathic surgery,