Imaging Science in Dentistry 2021; 51: 455-60
https://doi.org/10.5624/isd.20210046

An unusual presentation of osteoblastoma of the maxilla: A case report

Joonbum Lim[1], Nicole Hinchy[1], Nora Odingo[2], Dan Colosi[1], Mina Mahdian[1,*]

[1]*Division of Diagnostic Imaging, Department of Prosthodontics and Digital Technology, School of Dental Medicine, State University of New York at Stony Brook, New York, USA*
[2]*Division of Oral and Maxillofacial Pathology, Department of Oral and Maxillofacial Surgery, School of Dental Medicine, State University of New York at Stony Brook, New York, USA*

ABSTRACT

This report presents a rare case of maxillary osteoblastoma in a 17-year-old female. The patient presented with dull pain and facial asymmetry inferior to the left zygoma. An intraoral examination found a painless swelling on the buccal gingival tissue in the left posterior maxilla. Panoramic radiographs and multidetector computed tomographic images revealed an ill-defined, non-corticated, mixed attenuating entity of osseous density located within the left posterior maxilla apical to the left maxillary molars. The entity exhibited a heterogeneous internal structure with a fine granular appearance, and the periphery showed a partial hypo-attenuating rim along the antero-medial aspect. Expansion of the left posterior maxilla accompanied with displacement of the left maxillary sinus floor was noted. External root resorption of the first and second molars was noted, as well as postero-superior displacement of the third molar. The histopathologic diagnosis of the biopsy was osteoblastoma. Complete excision of the tumor was performed. ***(Imaging Sci Dent 2021; 51: 455-60)***

KEY WORDS: Osteoblastoma; Maxilla; Multidetector Computed Tomography; Radiography, Panoramic

Osteoblastoma is a rare benign neoplasm of osteoblasts, accounting for 1% of all primary bone neoplasms.[1] It is characterized by the proliferation of osteoblasts depositing osteoid and immature bone within a well-vascularized fibrous connective tissue stroma.[2] Microscopically, it exhibits central areas of irregular osteoid or woven bone rimmed by numerous osteoblasts and scattered osteoclasts, surrounded by a loose fibrovascular stroma.[3,4] The osteoblasts have ample cytoplasm and hyperchromatic nuclei, and the loose fibrous stroma includes dilated vessels and occasional hemorrhage.[3]

Osteoblastoma most commonly occurs in the second and third decades of life, with a male-to-female ratio of 2 : 1.[5,6] Although osteoblastoma can arise in any bone of the skeleton, it most often involves the long bones and vertebral column, with approximately 10% of cases found in the maxillofacial bones.[4] Most maxillofacial cases develop in the mandible (71.4%), especially in the posterior region.[2] Clinically, the most common symptoms are pain, swelling, warmth and tenderness of the affected region.[4,5] The pain is usually described as dull, aching, and often progressive in intensity.[4] The pain usually does not respond to non-steroidal anti-inflammatory drugs (NSAIDs) and is not generally more severe at night.[7]

The radiographic features of osteoblastoma have not been well established due to the lack of a comprehensive description of the radiographic findings.[8] In general, the radiographic features of osteoblastoma include a well- or ill-defined, round or oval-shaped entity with or without cortication that may include a radiolucent rim surrounding the more central areas of abnormal bone deposition.[3,5] The internal structure may have a completely radiolucent appearance, or it may show a mixed radiolucent/radiopaque appearance.[2,4,5] When a radiopaque center is present, the internal calcification may take the form of fine granular bone trabeculae.[5] The lesion can expand bone; the outer cortex, however, is usually maintained.[5] Displacement and/or resorption of adjacent

Received February 26, 2021; Revised May 25, 2021; Accepted May 28, 2021
Published online Aug 11, 2021
*Correspondence to : Dr. Mina Mahdian
Department of Prosthodontics and Digital Technology, School of Dental Medicine, State University of New York at Stony Brook, 139 Westchester Hall, South Drive, Stony Brook, NY 11794, USA
Tel) 1-631-632-8858, E-mail) mahdianmina@gmail.com

Imaging Science in Dentistry · pISSN 2233-7822 eISSN 2233-7830

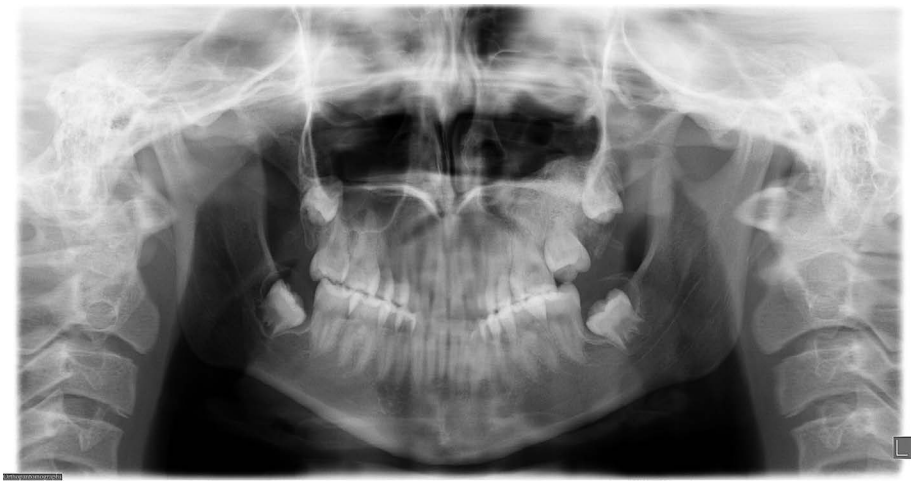

Fig. 1. Panoramic radiograph shows an ill-defined radiopacity without a peripheral radiolucent capsule involving the alveolar bone of the left posterior maxilla. Note displacement of the left maxillary sinus floor and the developing third molar.

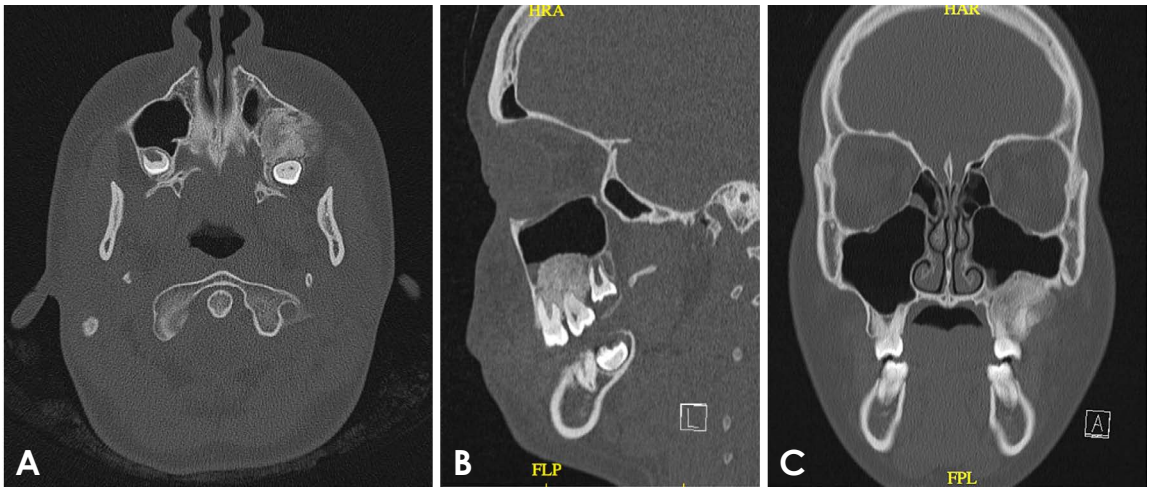

Fig. 2. A. Axial computed tomography (CT) image shows an ill-defined area of hyper-attenuation with a ground-glass appearance involving the left maxilla. Note a thin hypo-attenuating band at the medial periphery. B. Sagittal CT image shows an ill-defined area of hyper-attenuation with a ground-glass appearance involving the left maxilla. Note the directional external root resorption of the maxillary first and second molars, and superior displacement of the left maxillary sinus floor. C. Coronal CT image shows an ill-defined area of hyper-attenuation with a ground-glass appearance involving the left maxilla. A thin hypo-attenuating band at the medial periphery is also appreciated. Note the directional external root resorption of the maxillary first molar, and superior displacement of the left maxillary sinus floor.

teeth may also be seen.[5] This report presents a rare case of osteoblastoma involving the maxilla with radiographic features mimicking those of fibrous dysplasia of the jaw.

Case Report

A 17-year-old Hispanic female was admitted to the Stony Brook University School of Dental Medicine for an emergency visit with a chief complaint of dull pain of the left posterior maxilla. Physical examination found a bony swelling located buccal to the left maxillary molars. No erythema, edema, or purulent discharge was noted. Panoramic radiography revealed a moderately defined granular radiopaque area in the periapical region of the left maxillary molars with superior displacement of the sinus floor (Fig. 1). No peripheral radiolucent rim was noted on the panoramic radiograph. For further investigation of the lesion, bone algorithm multidetector computed tomography (MDCT) was performed. The MDCT images also revealed a moderately defined granular hyper-attenuating area in the region of the left maxillary molars. Superior displacement of the left maxillary sinus floor, supero-posterior displacement of the left maxillary third molar, and external apical root resorption of the first and second molars were also appreciated. The entity was confluent with the lamina dura of the affected teeth; however, the periodontal ligament

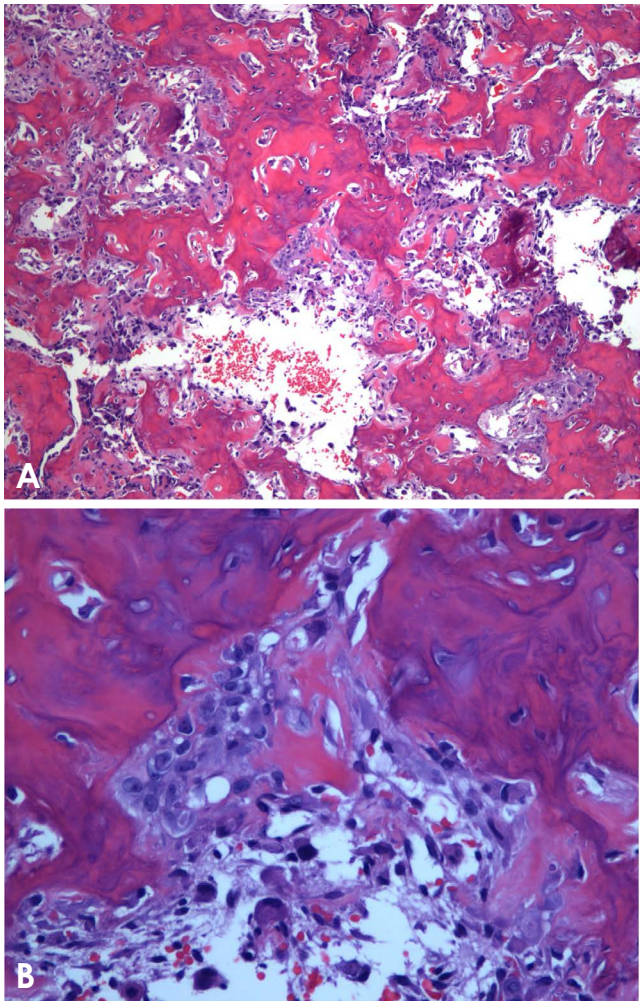

Fig. 3. A. Histopathologic findings of the incisional biopsy specimen. The lesion consists of well-vascularized, cellular fibrous connective tissue containing immature bone trabeculae surrounded by osteoblasts (H&E stain, original magnification ×100). B. Histopathologic findings of the incisional biopsy specimen. Note osteoblastic rimming and scattered osteoclasts surrounding trabeculae (H&E stain, original magnification ×400).

space remained intact. Of note were a thin hypo-attenuating rim, partially surrounding the central portion of the lesion at the antero-medial periphery, and a small area of possible buccal dehiscence adjacent to the left maxillary second molar (Fig. 2). Based on clinical and radiographic features, a preliminary differential diagnosis of osteoblastoma, fibrous dysplasia and cemento-ossifying fibroma was considered. Although unlikely, a remote possibility of osteosarcoma was considered as well. The patient, however, failed to attend her follow-up appointment for further examination.

The patient was then referred back to the authors' institution from a private oral and maxillofacial surgery office 13 months after the initial visit. An incisional biopsy had been performed by the referring physician and a histopathologic examination was done prior to readmittance. A histopathologic examination of the specimen revealed well-vascularized fibrous connective tissue containing a tumor composed of osteoid trabeculae surrounded by plump osteoblasts and scattered osteoclasts (Fig. 3). Osteoblast bridges between the bone trabeculae in several areas were also noted. Based on these features, a histolpathologic diagnosis of osteoblastoma was made.

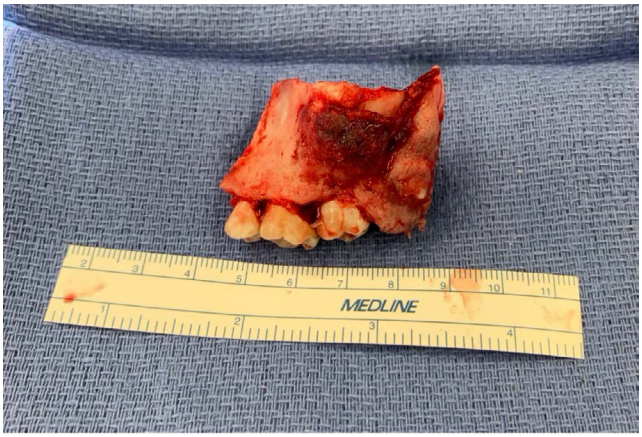

Fig. 4. Photograph of the gross specimen.

Complete excision of the tumor was performed via left partial maxillectomy with localized mucoperiosteal advancement flap under general anesthesia (Fig. 4). A histopathologic examination of the surgical specimen showed the histopathologic features of the previous incisional biopsy, and final diagnosis of osteoblastoma was made. The postoperative course was uneventful other than postoperative pain and nasal congestion, which eventually resolved by 4 weeks postoperatively. The patient was last seen 1 year after surgery and showed no sign of recurrence on both clinical and radiologic examinations (Fig. 5).

Discussion

Osteoblastoma was first reported by Jaffe and Mayer in 1932, when they reported a case of "an osteoblastic and osteoid tissue forming tumor" of a metacarpal in a 12-year-old female patient.[9] In 1956, Jaffe and Lichtenstein independently proposed the term "benign osteoblastoma" to designate "a rather vascular, osteoid- and bone-forming benign tumor characterized by the abundant presence of osteoblasts."[10,11] Osteoblastoma is a rare bone-forming tumor that rarely involves the maxillofacial bone, particularly the maxilla. Diagnosing osteoblastoma of maxillofacial bones

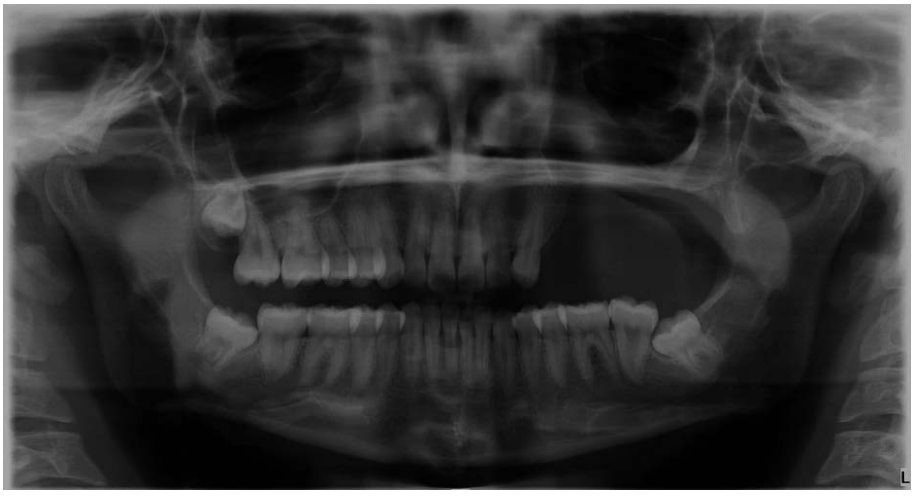

Fig. 5. Panoramic radiograph of the patient 1-year postoperatively reveals a surgical defect in the left posterior maxilla with clear margins. No sign of recurrence is noted.

can be challenging due to its rarity, variable clinical presentations, and overlapping radiographic and histopathologic features with other fibro-osseous lesions, benign tumors, and low-grade osteosarcoma. For this reason, it is very difficult, if not impossible, to differentiate osteoblastoma from other lesions considered in the differential diagnosis based solely on a single examination modality. Therefore, it is imperative to establish the diagnosis from the features of clinical, radiologic and histopathologic examinations.

This report presented a rare case of osteoblastoma in the maxilla with a radiographic appearance of ill-defined, fine granular hyper-attenuation with mild expansion of the involved bone elevating the maxillary sinus floor, resembling the radiographic features of fibrous dysplasia. This finding is in accordance with a previous report by Jones et al.,[2] where a case of osteoblastoma in the maxilla demonstrated ground-glass opacification. In addition, the preoperative differential diagnosis for 3 of the 5 cases of osteoblastoma in the maxilla in their report included fibrous dysplasia, further suggesting that osteoblastoma of the maxilla may resemble the radiographic characteristics of fibro-osseous lesions. Similar radiographic appearances were also seen in cases reported by Ohkubo et al.[11] and Shah et al.[8] Patients in both cases presented with ill-defined radiopaque expansile lesions involving the alveolar bone of the left posterior maxilla, resembling fibrous dysplasia. In this case, the radiographic appearance on panoramic radiography that differentiated osteoblastoma from fibrous dysplasia was extensive directional external root resorption of the involved teeth, which is a rare occurrence in fibrous dysplasia.

Despite subtle differences in radiographic features between osteoblastoma of the maxilla and fibrous dysplasia, the typical clinical and histopathologic features of osteoblastoma are different from those of fibrous dysplasia, and can be used to differentiate the 2 lesions. Clinically, patients with osteoblastoma of the jaw present with swelling of the affected area, and dull, aching pain that does not respond to NSAIDs.[2] A previous study by Jones et al.[2] found that approximately 70% of patients with osteoblastoma presented with pain, tenderness, and/or discomfort. While one of the most common clinical findings is swelling of the affected area, as in osteoblastoma of the jaw, pain is rare with patients with fibrous dysplasia.[5,12] In addition to the characteristic pain in the majority of cases of osteoblastoma, differences in histopathologic features may also aid in differentiating the 2 lesions. Microscopically, osteoblastoma shows broad seams of irregular trabeculae of osteoid or woven bone surrounded by numerous osteoblasts and scattered osteoblasts, set within a loose fibrovascular stroma.[3,4,13] The loose fibrous stroma exhibits greater vascularity, and occasional hemorrhage may occur. Of note is the osteoblast rimming, which is usually absent or minimal in case of fibrous dysplasia.[3] Therefore, osteoblastoma of the maxilla with an atypical radiographic appearance resembling fibrous dysplasia may be differentiated from fibrous dysplasia based on clinical and histopathologic features.

Given the ill-defined radiopacity without a peripheral radiolucent capsule on the initial panoramic radiograph, the initial differential diagnosis favored osteoblastoma and fibrous dysplasia over cemento-ossifying fibroma. The MDCT examination, however, revealed a thin hypo-attenuating rim, partially surrounding the antero-medial portion of the central area of fine granular bone deposition (Fig. 2A and C). Due to this well-defined medial periphery with a partially present hypo-attenuating rim, the possibility of cemento-ossifying fibroma was also considered. Cemento-

ossifying fibroma, a benign osseous neoplasm of the jaw, closely resembles many of the clinical, radiographic and histopathologic features of osteoblastoma. Radiographically, both lesions show a wide spectrum of features depending on the stage and amount of calcification. Other than the tendency of cemento-ossifying fibroma to have a well-defined periphery, the internal appearance of both lesions may present on a spectrum from complete radiolucency to complete radiopacity.[2,14,15] In addition, both lesions show tumor-like behavior (i.e., concentric expansion that usually leaves outer cortices of the affected bone intact, and displacement of adjacent structures and/or teeth).[5] Both lesions also have aggressive variants - namely, juvenile cemento-ossifying fibroma and aggressive osteoblastoma - that may cause erosion of adjacent osseous structures and/or teeth.[16] In fact, in the present case, the remote possibility of juvenile cemento-ossifying fibroma was considered due to the prominent external resorption of teeth in the affected area. Thus, it is extremely difficult, if not impossible, to differentiate osteoblastoma from juvenile cemento-ossifying fibroma based on radiographic features alone. The clinical presentation of both lesions involves swelling of the affected area, resulting in facial asymmetry. However, similar to fibrous dysplasia, patients presenting with cemento-ossifying fibroma are usually asymptomatic.[3,5] Triantafillidou et al.,[17] in their case report and the review of the literature, showed that only 1 of 14 patients (7%) with cemento-ossifying fibroma presented with pain. While approximately half of the patients with osteoblastoma were asymptomatic, approximately 70% of patients presented with pain that did not respond to NSAIDs.[2] Microscopically, the central feature that distinguishes osteoblastoma from cemento-ossifying fibroma is the lack of cellular spindle cells in the stroma; instead, osteoblastoma usually presents with a loose vascular stroma with numerous prominent epithelioid-type osteoblasts.[12]

Osteosarcoma is another lesion that must be differentiated from osteoblastoma. A well-differentiated osteosarcoma may be difficult to distinguish from osteoblastoma.[18] In particular, a rare form of osteosarcoma known as osteoblastoma-like osteosarcoma shares radiographic features with osteoblastoma, posing a further challenge for distinguishing it from osteoblastoma.[19] However, osteosarcoma usually shows more aggressive behavior, such as destruction of cortical bone, tooth resorption, and/or invasion into adjacent soft tissue.[5,20] A sunburst radiographic appearance may also be seen in osteosarcoma.[20] Moreover, osteoblastoma can be differentiated from osteosarcoma based on histopathologic features; osteosarcoma usually demonstrates cellular pleomorphism, high mitotic rate and/or tumor giant cells.[2,5] Despite these radiographic and histopathologic features that help differentiate osteoblastoma from osteosarcoma, the possibility of malignant transformation of osteoblastoma should not be overlooked, although it is a rare occurrence.[21] In 2017, Salmen and colleagues[22] described an aggressive case of maxillary osteoblastoma that relapsed following initial enucleation. They reported that cortical erosion and invasion into the adjacent structures is a rare and unusual behavior of the aggressive variant of osteoblastoma, resulting in the consideration of osteosarcoma in the differential diagnosis and requiring *en bloc* resection.

Other less frequently reported entities resembling osteoblastoma include cementoblastoma and osteoma. Cementoblastoma and osteoblastoma are proposed to be similar entities, differentiated only by their anatomic location; cementoblastomas occur exclusively in tooth-bearing areas. Radiographically, cementoblastoma has a distinct radiolucent halo, separating the entity from the adjacent bone, and the lesion is typically centered on and is continuous with the tooth structure. Osteoblastoma lacks a radiolucent halo with a center in the alveolar process.[23] Bilodeau et al.[23] reported a case of mandibular cementoblastoma with histologic and radiographic features mimicking osteoblastoma and osteosarcoma. McCann et al.[24] reported a case of craniofacial osteoma presenting with pain in the orbito-frontal region resembling sinusitis. Radiographically, the lesion appeared as a mixed radiodensity expansile entity with ground-glass fibro-osseous-like features in the frontal sinus, closely resembling the features of the present case, and the final histologic diagnosis was osteoblastoma with osteoblastoma-like features.

Last of all, one of the most closely related lesions to osteoblastoma, in terms of both radiographic and histologic features, is osteoid osteoma. Their radiographic features are similar, if not identical; the only difference is the size. Osteoid osteoma is smaller, typically less than 2 cm in diameter, whereas osteoblastoma is usually larger than 2 cm with an average size of 3.5 to 4 cm.[4] Clinically, the most common chief complaint is swelling or a firm, palpable mass, which is found in approximately 90% of patients with osteoblastoma.[17] In addition, patients often present with dull, aching pain that neither responds to NSAIDs nor becomes more severe at night.[2,11] Jones et al.[2] found that approximately 70% of patients with osteoblastoma presented with pain, tenderness, and/or discomfort. These two features - pain that does not become progressively severe at night, and pain that is irresponsive to NSAIDs - may differentiate osteoblastoma from osteoid osteoma, beside their

size difference.

In conclusion, the clinical, radiographic, and histopathologic findings of a case of osteoblastoma of the maxilla were presented in this case report. The findings of the present case suggest that osteoblastoma involving the maxilla may show a ground-glass or orange-peel appearance in the central area of abnormal bone deposition without a peripheral radiolucent rim, instead of the fine granular appearance that is more commonly seen in osteoblastoma. Due to its relatively rare occurrence in the jaw and the overlapping characteristics with other more common lesions, diagnosis of this rare tumor must be established based on a combination of clinical, radiographic, and histopathologic examinations. Finally, one should always keep in mind that osteoblastoma of the maxilla may show an atypical radiographic appearance and may resemble fibro-osseous lesions such as fibrous dysplasia.

Acknowledgements

We thank Dr. Peter Balacky for providing us with the details on the treatment course and the photograph of the gross surgical specimen. We also thank Dr. Renee Reich for providing us with the photomicrographs of the incisional biopsy specimen.

Conflicts of Interest: None

References

1. MacDonald D. Oral and maxillofacial radiology: a diagnostic approach. 2nd ed. Chichester: Wiley-Blackwell; 2020. p. 317-9.
2. Jones AC, Prihoda TJ, Kacher JE, Odingo NA, Freedman PD. Osteoblastoma of the maxilla and mandible: a report of 24 cases, review of the literature, and discussion of its relationship to osteoid osteoma of the jaws. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006; 102: 639-50.
3. Neville BW, Damm DD, Allen CM, Chi AC. Oral and maxillofacial pathology. 4th ed. St. Louis: Elsevier; 2016. p. 608-9.
4. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg 2011; 19: 678-89.
5. Mallya SM, Lam EW. White and Pharoah's oral radiology: principles and interpretation. 8th ed. St. Louis: Elsevier; 2019. p. 442-3.
6. Angiero F, Mellone P, Baldi A, Stefani M. Osteoblastoma of the jaw: report of two cases and review of the literature. In Vivo 2006; 20: 665-70.
7. Berry M, Mankin H, Gebhardt M, Rosenberg A, Hornicek F. Osteoblastoma: a 30-year study of 99 cases. J Surg Oncol 2008; 98: 179-83.
8. Shah S, Kim JE, Huh KH, Yi WJ, Heo MS, Lee SS. Recurrent osteoblastoma of the maxilla. Dentomaxillofac Radiol 2013; 42: 20100263.
9. Jaffe HL, Mayer L. An osteoblastic osteoid tissue-forming tumor of a metacarpal bone. Arch Surg 1932; 24: 550-64.
10. Jaffe HL. Benign osteoblastoma. Bull Hosp Joint Dis 1956; 17: 141-51.
11. Ohkubo T, Hernandez JC, Ooya K, Krutchkoff D. "Aggressive" osteoblastoma of the maxilla. Oral Surg Oral Med Oral Pathol 1989; 68: 69-73.
12. MacDonald-Jankowski D. Fibrous dysplasia in the jaws of a Hong Kong population: radiographic presentation and systematic review. Dentomaxillofac Radiol 1999; 28: 195-202.
13. McCarthy EF. Fibro-osseous lesions of the maxillofacial bones. Head Neck Pathol 2013; 7: 5-10.
14. MacDonald-Jankowski DS. Cemento-ossifying fibromas in the jaws of Hong Kong Chinese. Dentomaxillofac Radiol 1998; 27: 298-304.
15. MacDonald-Jankowski. Fibro-osseous lesions of the face and jaws. Clin Radiol 2004; 59: 11-25.
16. Slootweg PJ, Panders AK, Koopmans R, Nikkels PG. Juvenile ossifying fibroma. An analysis of 33 cases with emphasis on histopathological aspects. J Oral Pathol Med 1994; 23: 385-8.
17. Triantafillidou K, Venetis G, Karakinaris G, Iordanidis F. Ossifying fibroma of the jaws: a clinical study of 14 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol 2012; 114:193-9.
18. Koenig LJ, Tamimi D, Petrikowski CG, Perschbacher SE. Diagnostic Imaging: Oral and Maxillofacial. 2nd ed. Philadelphia: Elsevier; 2018. p. 478-9.
19. Ozger H, Alpan B, Söylemez MS, Ozkan K, Salduz A, Bilgic B, et al. Clinical management of a challenging malignancy, osteoblastoma-like osteosarcoma: a report of four cases and a review of the literature. Ther Clin Risk Manag 2016; 12: 1261-70.
20. Bilodeau E, Collins B, Costello B, Potluri A. Case report: a pediatric case of cementoblastoma with histologic and radiographic features of an osteoblastoma and osteosarcoma. Head Neck Pathol 2010; 4: 324-8.
21. Woźniak AW, Nowaczyk MT, Osmola K, Golusinski W. Malignant transformation of an osteoblastoma of the mandible: case report and review of the literature. Eur Arch Otorhinolaryngol 2010; 267: 845-9.
22. Salmen FS, Oliveira MR, Navarro CM, Dedivitis RA, Pereira Filho VA, Gabrielli MF. Aggressive osteoblastoma in the maxilla: unusual lesion in the craniofacial skeleton. J Craniofac Surg 2017; 28: 794-7.
23. Bilodeau E, Collins B, Costello B, Potluri A. Case report: a pediatric case of cementoblastoma with histologic and radiographic features of an osteoblastoma and osteosarcoma. Head Neck Pathol 2010; 4: 324-8.
24. McCann JM, Tyler D Jr, Foss RD. Sino-orbital osteoma with osteoblastoma-like features. Head Neck Pathol 2015; 9: 503-6.