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Case report

Improperly diagnosed odontogenic myxoma in a 23-year-old female: A radiographic analysis

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ABSTRACT

Background: *Misdiagnosis can occur due to various radiographic alterations linked to odontogenic myxoma (OM). Regular examination can detect abnormalities early on, but not all practitioners are aware that these lesions exist.* ***Purpose:*** *This case report aims to describe and discuss an OM case from the perspective of oral radiology on panoramic radiographs and cone-beam computed tomography (CBCT).* ***Case:*** *A 23-year-old female went to her first dentist for orthodontic treatment with no prior radiographic evaluation. On January 7th, 2022, the second dentist extracted teeth 38 and 48 using the panoramic radiograph without identifying lesions. Concerned about swelling on her lower right gingiva, which had gradually grown, the patient went to an oral and maxillofacial surgeon on November 15th, 2022. The clinical examination revealed facial asymmetry with a thick, palpable, firm mass with an ambiguous boundary. Despite the evident movement of tooth 47, the gingiva exhibited no noticeable change in coloration.* ***Case management:*** *From the panoramic examination, multilocular radiolucency with radiopaque septa and aggressive mass characteristics were found. Advanced imaging CBCT was used to investigate further and correlate histology findings for treatment.* ***Conclusion:*** *Odontogenic myxoma is difficult to distinguish from other benign and malignant neoplasms due to the wide variations of radiological patterns. Cone-beam computed tomography provides a thorough and broad range of data that can be used to make a precise diagnosis and develop an effective treatment strategy. This highlights the critical need for a trained expert to thoroughly examine CBCT scans.*

Keywords: *CBCT; odontogenic myxoma; panoramic; tumor benign*

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INTRODUCTION

Odontogenic myxoma (OM) is a benign intraosseous tumor that develops from odontogenic ectomesenchyme and resembles mesenchymal components of the dental papilla. Comprising 3% to 6% of odontogenic tumors, these lesions are incredibly uncommon. While this OM does not metastasize, it is poorly encapsulated and has a propensity to invade the cancellous bone in the area. These lesions are slow-growing and harmless, but if left untreated, they can enlarge and cause facial asymmetry. This odontogenic tumor has a predilection for occurring in the posterior regions of both the mandible and maxilla, with a higher incidence observed in women than in men.[1,2]

The recurrence rate is quite high—up to 25%—since this form of tumor is not encapsulated. Therefore, the boundary of the lesion is not evident (poorly defined), and there are jelly-like myxoid sacs that can nest in the trabecular bone cavities, making excising the lesion difficult. Bone trapped within the lesion goes through remodeling, becoming bent and straight to create septa, giving the appearance of being multilocular. This indicates that septal characteristics in OM, such as a tennis racket-like or stepladder-like pattern, may aid in identifying the lesion.[1,3]

Loose teeth are frequently seen as a common dental condition, even though there are numerous etiologies for tooth mobility, which can be physiological or pathological. Dentists must identify the causal sources, including in this case where tooth extraction was immediately performed without careful examination.[4] Odontogenic myxoma exhibits varied radiographic features that can often lead to establishing a misleading diagnosis of an OM border being

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either well-defined or ill-defined.[5] Making a diagnosis as a clinician is crucial since it affects both patients and other medical professionals. The diagnostic procedure entails gathering information from the patient's medical history, conducting a clinical examination, administering diagnostic tests, and interpreting and integrating the results to determine the patient's diagnosis. The importance of interpreting and visually inspecting radiography results is comparable.[6] This case report aims to describe and discuss a case from the perspective of oral radiology, as determined by panoramic radiographs and cone-beam computed tomography (CBCT).

CASE

A 23-year-old female went to her first dentist for orthodontic treatment with no prior radiographic evaluation. Due to worries that the impacted tooth could interfere with therapy, the patient was referred for a panoramic examination on January 7th, 2022 while undergoing orthodontic treatment. The second dentist performed an odontectomy on teeth 38 and 48 using the existing panoramic radiograph without identifying any lesions. There had been concerns afterward regarding the progressive enlargement of the lower right gums, which contributed to facial asymmetry. On November 15th, 2022, the patient went to an oral and maxillofacial surgeon and was referred for a panoramic examination. A lesion in the right mandible was suspected. To find out more about the characteristics of the lesion and plan therapy, she was referred for a CBCT examination.

Figure 1. Clinical intra-oral examination.

Clinical examination (Figure 1) revealed facial asymmetry and painless enlargement on the right lower gingiva with an indistinct boundary that felt thick and solid to palpation. There was tooth mobility on teeth 47, and the color of the gingiva was normal.

The first panoramic radiographic examination (Figure 2) was carried out before the odontectomy was performed in January. It revealed radiopaque, visibly fixed orthodontic equipment and impacted teeth 38 and 48; unaware of the lesion, the second dentist performed an odontectomy on both affected teeth.

A panoramic examination (Figure 3) was performed again because the patient complained of loose tooth 47 and gum expansion. An oral and maxillofacial surgeon was aware of the lesion and that there was a multilocular radiolucency with radiopaque septa similar to an ameloblastoma. Because the picture obtained was less typical and other malignant lesions were suspected, a CBCT examination was carried out.

A CBCT examination was performed to obtain a more detailed and comprehensive radiograph. A reconstructed CBCT examination of the maxilla and mandible was performed with a field of view of 9 x 14. A semi-ovoid, irregular, radiolucent lesion with scalloped margins extended from the body (region 46) to the dextra mandibular ramus involving tooth 47 and was accompanied by mandibular inferior cortical thinning.

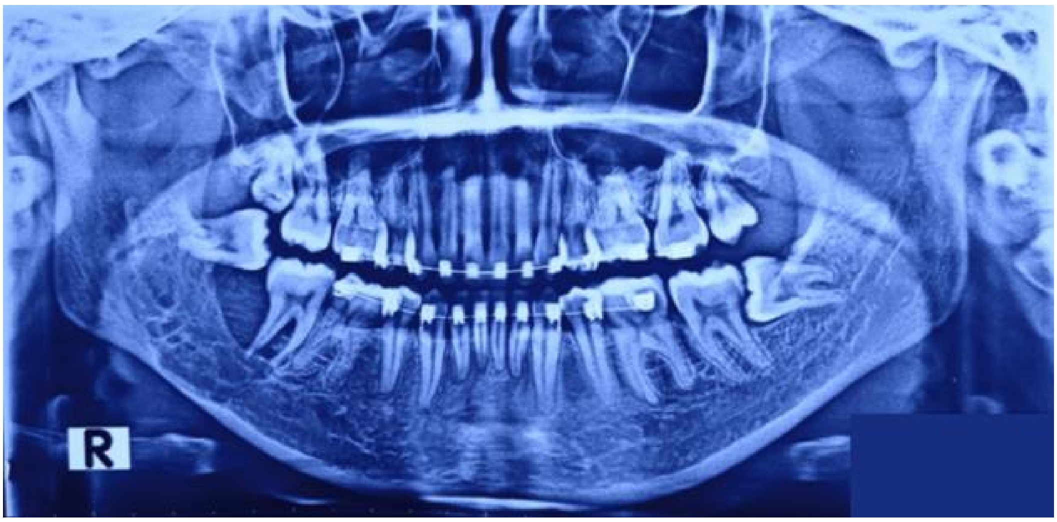

Figure 2. The first panoramic examination was on January 7th, 2022.

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Figure 3. The second panoramic examination was on November 15th, 2022.

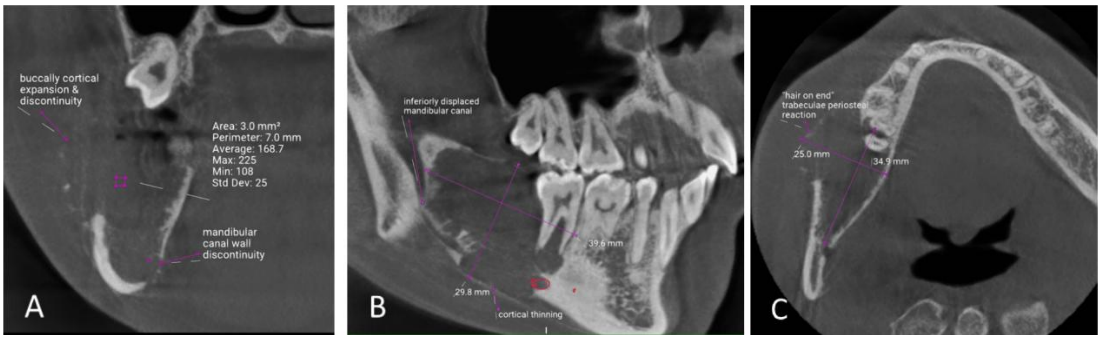

Figure 4. Multiplanar reconstruction: A. Coronal view; B. Sagittal view; C. Axial view.

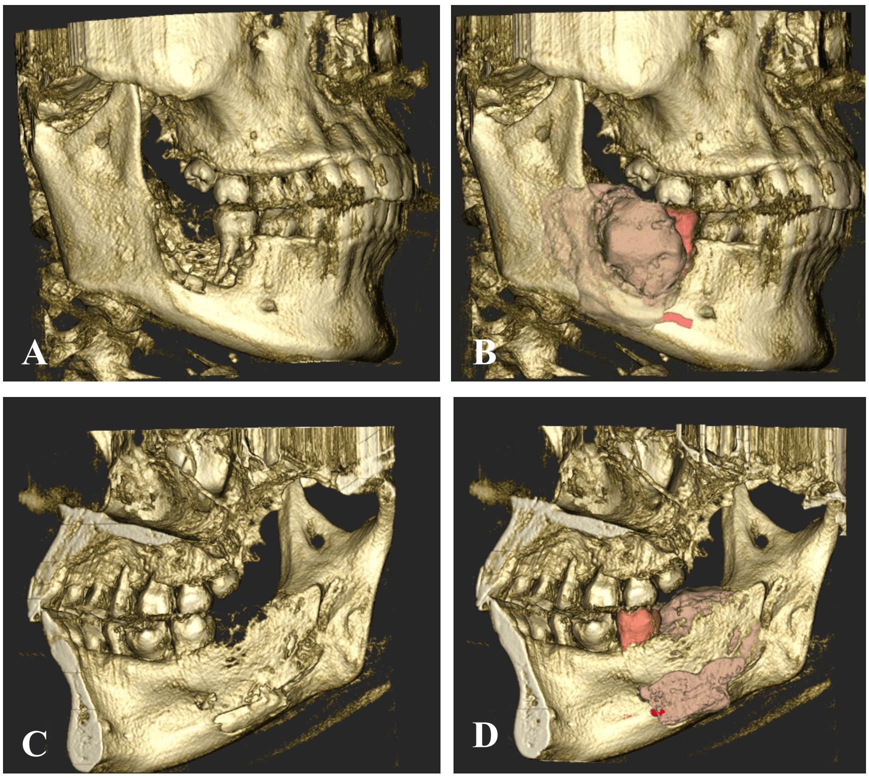

Figure 5. Reconstructed CBCT 3D view and 3D view with segmentation (A and B buccal view; C and D lingual view).

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CASE MANAGEMENT

CBCT reconstruction (Figure 4) showed a semi-ovoid or circular, unilocular, radiolucent mass with largely well-defined and a few ill-defined edges running from the body mandible of region 46 to the ramus of the mandible. The internal structure of the lesion (Figure 4A) showed a density with an average region of interest of 168.7 gray values, similar to soft tissue or fluid mass density. The lesion expanded with thinning and cortical perforation of the mandible on the lingual inferior and buccal sides (pink arrows). Perforations in the buccal cortical and enlarged cortical bones remained to provide the impression of a hair-on-end pattern, raising the possibility of an aggressive lesion.

The sagittal section of the lesion (Figure 4B) measured approximately 29.8 x 39.6 x 25 mm. This slice depicted a lesion involving the mandibular canal, pushing the canal inferiorly and eliminating the cortical wall of the canal in the middle of the lesion.

The axial slice (Figure 4C) depicted the lesion expanding to the anteroposterior and buccolingual regions as well as the thinning and perforation of the buccal and lingual cortical plates (pink arrows). Lesions are typically expansive in a buccal direction with signs of cortical discontinuity. The plate perforation on the buccal side seemed to demolish bone with the appearance of a hair-on-end pattern, which is characteristic of an aggressive lesion.

A reconstructed 3D view with segmentation (Figure 5) can demonstrate the dimensions of the lesion for better visualization. The mandibular canal was displaced inferiorly, accompanied by a loss of the cortical wall or a discontinuity in the middle of the corpus mandible. The lesion affected both the apical and furcation of tooth 47, and there was no resorption or morphological alteration in the teeth.

To confirm the diagnosis, a histological examination was undertaken. The macroscopic images received were fragments of biopsy tissue with a total size of 15 x 15 x 5 mm, a gray-white color, and a soft, solid consistency processed in two cassettes. The microscopic view (Figure 6) shows tissue sections made of myxoid material with stellate cells, with the solid component made of spindle cells grouped in irregular bundles. There were no abnormal cells or tumors. The results of the investigation point to an OM.

DISCUSSION

Detection and discrimination of pathological states in a radiographic image requires a perceptual process, which involves recognizing variations in existing images, and a cognitive process, which involves comprehending the substantial alterations that occur. The clinician must be able to detect and define pathological abnormalities to make a precise diagnosis. This is a complicated process, especially when decisions based on limited information result in a diagnosis error. Hedge et al. stated that several factors, including the complexity of the radiographic images, clinical case experience, clinical and technical knowledge of radiographic examination, lack of conceptual understanding of knowledge, case resolution, limited time due to working hours and heavy workload, and a lack of training or educational programs, can all influence misinterpretation.[6]

Odontogenic myxoma is an intraosseous tumor of the jaw, and radiographic evaluation is required for diagnosis. It is interesting in this case because the failure of more than one dentist to notice the lesion resulted in its progression from asymptomatic to enlarged. The patient received orthodontic treatment without undergoing a preliminary radiographic evaluation. Throughout the treatment, no noticeable symptoms or complaints were seen until the eruption of the impacted wisdom teeth was perceived to impair the progress of the treatment. Panoramic dental radiographs are frequently used as a diagnostic tool before orthodontic treatment. The two-dimensional image is highly relevant in the field of medical examinations and diagnostics.

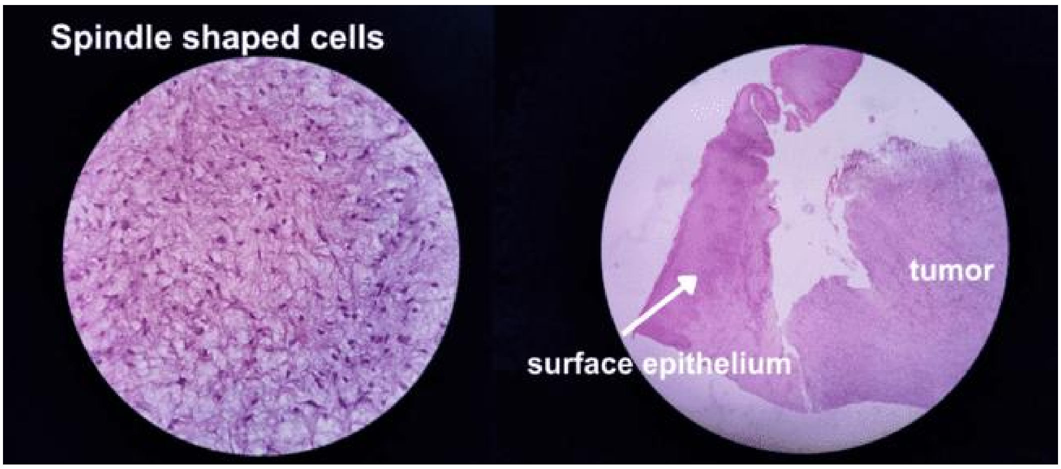

Figure 6. Result of histopathological examination.

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The tentative diagnosis in this case was ameloblastoma because it was clinically associated. Its radiography pictures indicated a benign, aggressive, odontogenic tumor with unilocular ameloblastoma as a differential diagnosis. A figure of expansion, a cortical perforation, and an ill-defined border indicated a hair-on-end pattern on some sides radiographically, so one could consider the possibility that the inflammatory lesion was aggressive and possibly malignant.[7] Based on the typical aggressive findings of the mass radiographically, the chance of a cystic lesion was modest, although it could still have been a differential diagnosis.

The radiographic appearance of the OM may have varied and, in this case, did not show the characteristic straight septa. Various radiographic features could be identified as OM (Figure 7). In the conventional radiographic examination, to facilitate identification, Zhang et al. (2007) divided OM into several radiographic figures, namely: Type I—unilocular with a radiolucent cavity; Type II—multilocular (including honeycomb, soap bubble, and tennis racket patterns); Type III—involvement with resorbed alveolar bone even if the lesion is small; Type IV—involvement with the maxillary sinus; Type V—osteolytic destruction with a pattern of large, irregularly demarcated radiolucent areas and erosion of cortical bone (moth-eaten borders); and Type VI—a mixture of osteolytic destruction and osteogenesis that has an internal radiopaque appearance and a sunray appearance.[5] Although OM is a benign tumor, in certain cases, it may also display a sunray or the sunburst appearance that usually represents some malignancy, such as osteosarcoma.[8]

The combination of detecting straight and sharp septa on radiography and orientation of an age-based approach, which predilect to occur in the third decade of life, can greatly aid in the diagnosis of OM.[5,12] The presence or absence of these lobules/septa also demonstrates tumor progression stages. Radiographic findings frequently reveal the presence of a lesion invasion into the bone cortex. Septa have been discovered to be reoriented cortical bone or

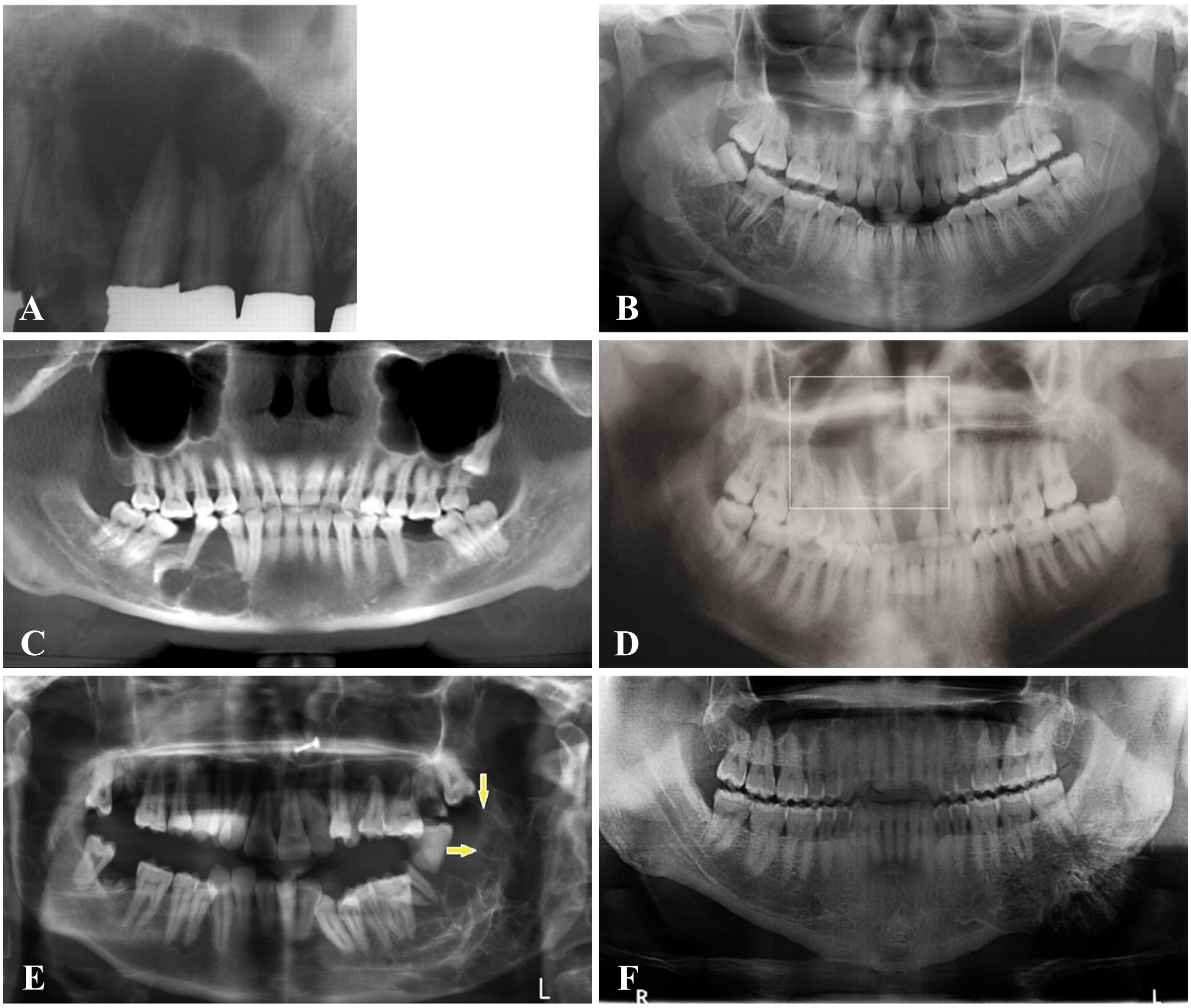

Figure 7. Several variations of the OM radiographic appearance; A. OM with unilocular radiolucent;[9] B. OM with radiolucency with fine, internal opaque trabeculations of the right posterior mandible;[10] C. OM with typical tennis racket appearance;[3] D. OM with the maxillary sinus involvement;[11] E. OM with a large, ill-defined multilocular radiolucency in the left mandibular body and ramus with a honeycomb or tennis racket appearance with straight septa along the periphery of the lesion (arrows);[7] F. OM with a sunray or sunburst pattern.[8]

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sheets of dense, fibrous connective tissues with ambiguous borders that resemble malignancy. The periosteal response functions as a barrier, preventing tumor growth into soft tissues. This can cause soft tissue compression to produce a pseudo capsule, allowing the tumor to be identified from surrounding tissue even in the lack of cortex.[4]

The differential diagnosis based on radiographic data varies according to the location of the lesion. As in a simple bony cyst, the unilocular OM lesion has a scallop boundary between the teeth involved. Because of its multilocular internal structure, OM has a differential diagnosis that includes osteosarcoma, odontogenic keratocyst, intraosseous hemangioma, and ameloblastoma.[1] Ameloblastoma has a spherical structure as opposed to OM, which has a square or triangular form where multiple cystic ameloblastomas can invade nearby tissues and have the potential to metastasize.[13] Compared to OM, an ameloblastoma's cortical border is more defined and less likely to penetrate soft tissue or damage cortical bone integrity. In comparison to intraosseous hemangiomas, they typically develop in people under the age of 30 and are most common in the anterior maxillary and mandibular regions. Hemangiomas can cause aberrant expansion of the mandibular canal, mental foramen, and mandibular foramen when they are connected with the mandibular canal. Clinical signs include gingival redness and easy bleeding.[14]

Tumors might be identified by chance during routine examinations, or they can cause symptoms such as pain, paresthesia, tooth displacement, tooth mobility, and external resorption.[4,15] In this particular case, there was a high likelihood of the dentist failing to interpret radiographic data and, therefore, failing to notice that there was a lesion surrounding tooth 47, which was experiencing movement related to impacted tooth 48. The morphology of septa, clear and expanding radiolucent areas, cortical bone thinning, and even mandibular cortical perforations on the inferior lingual and buccal sides demonstrate that while OM growth is generally slow, it can nonetheless be expansive.

Panoramic radiography is routinely requested preoperatively; however, due to the limits of a two-dimensional image and the occurrence of image distortions that may interfere with surgical planning, panoramic evaluation is not sufficient in many cases. Accurate examination of panoramic radiographs requires a basic understanding of human anatomy. Furthermore, it is critical to carefully evaluate and analyze the different normal variations and variations in lesion characteristics to avoid incorrect interpretations. To that aim, radiographic pictures should be reviewed carefully by a knowledgeable specialist.

Two-dimensional image limitations can be circumvented by using CBCT, which can prevent geometric distortion, superimpose anatomic structures, and display the interior structure of the lesion with more precision than traditional radiography.[10,16] Radiologically, there are notable distinctions between panoramic and CBCT images of intraosseous lesions related to the integrity of the corticated border, the impact on surrounding tissue, cortical thinning, and cortical bone destruction. The utilization of advanced imaging techniques holds potential for the analysis of the internal structure of lesions and the assessment of bone borders, especially the diagnosis of OM because of its soft tissue-invading features.[17] This evaluation can provide valuable insights into the nature of the lesion, facilitating the development of a treatment plan that aligns with histological findings. After surgery, CBCT proves useful for tracking lesions with high recurrences because of its accuracy in measuring, lack of image distortion, and lesion boundary accuracy.[18]

Histopathology reveals stellate, spindle-shaped cells with pale or slightly eosinophilic cytoplasm radiating from a central nucleus. Large numbers of cells are equally distributed in a mucoid or myxoid stroma that contains some fine collagen fibers.[19] The amount of myxoid and fibrous tissue components in an examination, as well as the level of cell polarization, results in distinct radiography findings.[5] The gelatinous composition of the myxoma allows the tumor to infiltrate through skeletal trabeculation, leaving indistinct boundaries and making removal difficult.[20] PET scan evaluation can confirm the presence of metastases, excluding the potential of cancer.[15]

Conservative enucleation treatments may not eliminate the lesion and are prone to recurrence, whereas surgical resection removes diseased tissue better. Because of its jelly-like substance and form, as well as the lack of a capsule, OM has a high recurrence rate, requiring 2 to 15 years of clinical and radiological monitoring.[21,22] Rather than the characteristics of the tumor, the rate of recurrence is related to the therapy techniques, size, location, removal of an unerupted tooth, and the patient's ability to be followed up with further treatment.[23–25] The surgical margins for radical excision are typically 1–2 cm apart until they reach normal bone with tumor-free margins. However, Takahashi et al. concluded that a prospective study is still necessary to figure out the optimum surgical margins for OM. Recurrence rates can be reduced with radical treatment choices; however, reconstructing surgical defects becomes incredibly challenging.[26]

The potential ramifications of misinterpretation can significantly impact the accuracy and efficacy of diagnostic and treatment determinations. The interpretation and differential diagnosis of OM are challenging due to the high overlap with other benign and malignant neoplasms resulting from the varied spectrum of radiographic patterns observed. Cone-beam computed tomography can provide a comprehensive and extensive set of data that can be utilized to make an accurate diagnosis and devise an efficient treatment plan and should be carefully reviewed by a trained professional.

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