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CASE REPORT / OLGU SUNUMU

Odontogenic Myxoma: Clinical and Radiographic Characteristics of Two Cases

Odontojenik Miksomanın Klinik ve Radyografik Özellikleri: İki Olgu Sunumu

Melda Pelin AKKİTAP, Birsay GÜMRÜ, Bilge TARÇIN, Ender İDMAN

Abstract

Odontogenic myxoma (OM) is a benign, slow-growing neoplasm of ectomesenchymal origin. The radiological features are variable including unilocular or multilocular radiolucency with a well-defined or diffuse border, making the differential diagnosis difficult. These facts pose potential challenges in reaching the correct diagnosis only with radiographic examinations. In this article, two cases of OM are presented and the varied clinical and radiological features and difficulties in differential diagnosis are discussed together with the application of computed tomography (CT) and cone beam computed tomography (CBCT).

The presented cases are a 33-year-old female patient with the complaints of migration and mobility of maxillary left premolar teeth and a 30-year-old-male patient with the complaint of a firm, non-tender, fixed mass in the mandibular right posterior region. Periapical, panoramic, and CT/CBCT images revealed the presence of multilocular hypodense lesions with or without hyperdense foci in the relevant areas, and divergence and mobility of the associated teeth were also observed. Excisional biopsy and histopathological examination exhibited the definitive diagnosis of OM.

Three-dimensional advanced imaging techniques such as CT and CBCT differ from two-dimensional periapical and panoramic radiographs in that they can demonstrate more characteristic radiographic findings and clearly display the tumour in three-dimensional accuracy and perspective. Therefore, the use of advanced imaging techniques is recommended as a useful tool in the diagnostic process of OM in order to display the borders and to help in investigation of the internal structure of the tumour.

Keywords: odontogenic myxoma, diagnosis, panoramic radiography, computed tomography, cone beam computed tomography

Öz

Odontojenik miksoma (OM), ektomezenkimal kökenli benign, yavaş büyüyen bir neoplazmadır. Sınırları belirgin veya difüz olan uniloküler veya multiloküler radyolüsent görüntü şeklinde değişkenlik gösteren radyolojik özellikleri ayırıcı tanısını zorlaştırmaktadır. Bu nedenle, sadece radyografik incelemelerle doğru tanıya ulaşmada güçlükler ortaya çıkmaktadır. Bu makalede iki OM olgusu sunulmakta ve değişkenlik gösteren klinik ve radyolojik özellikler ve ayırıcı tanıdaki zorluklar bilgisayarlı tomografi (BT) ve konik ışınlı bilgisayarlı tomografi (KIBT) uygulamaları ile birlikte tartışılmaktadır.

Sunulan vakalar; sol maksiller premolar dişlerinde yer değişikliği ve mobilite şikayeti ile başvuran 33 yaşında kadın hasta ve sağ mandibular posterior bölgesinde sert, ağrısız, hareketsiz kitle varlığı şikayeti olan 30 yaşında erkek hastadır. Periapikal, panoramik ve BT/KIBT görüntülerinde, söz konusu bölgelerde hiperdens odaklar içeren veya içermeyen multiloküler hipodens lezyonlar izlenmiştir ve ilgili dişlerde de yer değişikliği ve mobilite gözlenmiştir. Eksizyonel biyopsi ve histopatolojik inceleme sonucunda OM kesin tanısına ulaşılmıştır.

BT ve KIBT gibi üç boyutlu ileri görüntüleme teknikleri, daha karakteristik radyografik bulgular sağlayabilmeleri ve tümörü üç boyutlu doğruluk ve perspektifte net bir şekilde gösterebilmeleri açısından iki boyutlu periapikal ve panoramik radyografilerden farklıdır. Bu nedenle, sınırların görüntülenmesi ve tümörün iç yapısının değerlendirilmesinde yardımcı olmak için ileri görüntüleme tekniklerinin kullanılması OM'nin tanı sürecinde yararlı bir araç olarak önerilmektedir.

Anahtar Kelimeler: odontojenik miksoma, tanı, panoramik radyografi, bilgisayarlı tomografi, konik ışınlı bilgisayarlı tomografi

Birsay Gümrü (✉)
Marmara Üniversitesi, Diş Hekimliği Fakültesi, Ağız, Diş ve Çene Radyolojisi Anabilim Dalı, Başıbüyük Mah. Başıbüyük Yolu Sok. 9/3 34854 Başıbüyük, Maltepe/İstanbul
e-mail: bgumru@marmara.edu.tr

Melda Pelin Akkitap, Birsay Gümrü, Ender İdman
Marmara University Faculty of Dentistry, Department of Oral and Maxillofacial Radiology, Istanbul, Turkey

Bilge Tarçın
Marmara University Faculty of Dentistry, Department of Restorative Dentistry, Istanbul, Turkey

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Introduction

Odontogenic myxoma (OM) is an aggressive, locally invasive, and non-metastatic odontogenic benign tumour considered to derive from the embryonic mesenchymal components of dental anlage such as dental follicle, dental papilla or periodontal ligament (1-6). OM, which accounts for about 3-6% of all odontogenic tumours, is considered to be a non-encapsulated neoplasmic process presenting in the tooth-bearing areas of the maxilla and mandible (3-5, 7-9).

Since the first and original description by Thoma and Goldman in 1947 as an uncommon benign tumour arising in the tooth-bearing areas, the origin of OM is still controversial (6, 10). It is believed to originate from the ectomesenchyme of a developing tooth and/or the undifferentiated mesenchymal cells of the periodontal ligament (6, 11-13). The World Health Organization classified OM as "a benign tumour of ectomesenchymal origin with or without odontogenic epithelium" (3, 11, 14). The idea that the neoplasm is of odontogenic origin has arisen due to reasons such as the histological resemblance to pulpal ectomesenchyme, the particular development nearby the tooth-bearing areas of the maxilla and mandible, the decreased incidence in case of missing or impacted teeth, the existence of inactive odontogenic epithelium in very few cases, and the infrequent development in other skeletal regions of the human body (3, 12, 15).

The general rare clinical appearance, non-specific symptomatology, and various radiographic appearances of OM may lead to misdiagnosis and failure to recommend it as a potential pathology even in differential diagnosis (16).

In this article, the various radiographic features and the challenges in differential diagnosis of two OM cases are presented along with the clinical application of computed tomography (CT) and cone beam computed tomography (CBCT).

Case Presentation

Case 1

A 33-year-old female patient referred to the Oral and Maxillofacial Radiology Department of Dentistry Faculty in Marmara University, Istanbul, Turkey, with complaints of migration and mobility of maxillary left first and second premolars.

The medical and dental history of the patient was unremarkable. Extra-oral examination revealed no facial asymmetry. There were no signs of inflammation, such as erythema, elevated local temperature, or palpable regional lymph nodes. Intra-oral examination revealed no signs of pathology, but the teeth #24 and #25 were found to be mobile and desensitive.

The periapical radiograph and orthopantomograph (OPTG) demonstrated multilocular radioluceny with well-defined borders and non-sclerotic rim in the interradicular region of the teeth #24 and #25 and displacement of the mentioned teeth was detected (Figures 1a and b). CT images in the coronal, sagittal, and axial planes revealed the positional change of the teeth #24 and #25 and hypodense lesion of "soap bubble" pattern in the alveolar bone between mentioned teeth (Figure 1c).

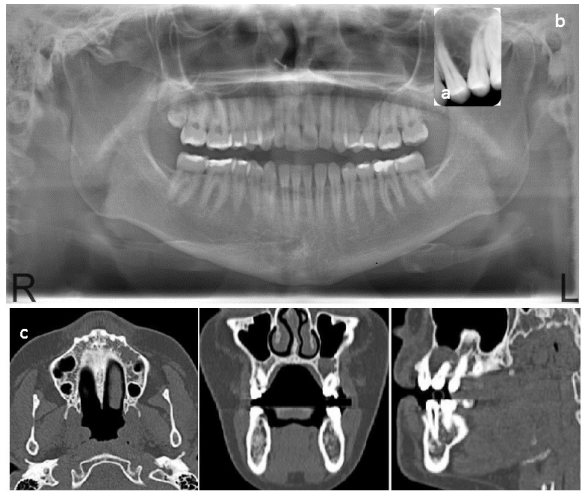

Figure 1: *(a) Periapical, (b) OPTG, and (c) CT (coronal, axial, sagittal planes) images of Case 1 showing multilocular radiolucent lesion in the interradicular region of maxillary left premolars.*

Following root-canal treatment of the teeth #24 and #25, excisional biopsy of the lesion was carried out under local anesthesia. The histopathological examination of the specimen revealed a benign lesion with features of OM.

Case 2

A 30-year-old male patient admitted to our clinic complaining of a pain-free swelling in the mandibular right posterior region.

His medical and dental histories were not contributory. In the extra-oral examination, a diffuse swelling in the mandibular right posterior region was noticed and the skin overlying the swelling appeared to be normal in terms of texture and colour. No increase in local temperature was detected and no signs of regional lymphadenopathy were evident. In the intra-oral examination, a firm, non-tender, fixed mass extending posteriorly from second premolar to the mandibular retromolar region was detected. The mucosa covering the area was intact. Clinical features were not specific and the related teeth were vital. Cross-bite of opposing teeth in the mandibular left posterior region was also observed (Figure 2).

Figure 2: *Intra-oral clinical presentation of Case 2.*

The OPTG revealed a multilocular radiolucent lesion reaching from the midline to the mandibular right ramus region exhibiting radiopaque foci, well-defined borders, and non-sclerotic rim. In addition, divergence of the teeth #45 and #46 was remarkable. The lesion was observed to scallop between the roots of the adjacent teeth (Figure 3a). The coronal, sagittal, and axial plane CBCT images demonstrated a well-defined hypodense lesion of approximately 58x30x34 mm, with hyperdense foci, bicortical destruction, and expansion with cortical perforation in the area of interest. In addition, the continuity of the mandibular nerve within the lesion was disrupted (Figure 3b).

Since the radiological examination was not able to provide satisfactory information for distinguishing the lesion, an incisional biopsy of the lesion was carried out under local anesthesia with diagnostic purposes. The histopathological examination of the specimen confirmed the diagnosis of OM.

Figure 3: *(a) OPTG and (b) CBCT (coronal, axial, sagittal planes) images of Case 2 showing multilocular radiolucent lesion of right mandible.*

Discussion

OM is a benign, slow-growing, and locally invasive neoplastic lesion, developing particularly in the tooth-bearing areas of the jaws (5, 14). The clinical findings, radiographic manifestations, and therapeutic approaches of OM have been documented by various case reports and studies in series. These studies show that OM occurs most frequently in patients from the second to the fourth decades, reaching the peak in the third decade of life, and the incidence decreases in those younger than 10 years and older than 50 years old (1-3, 8, 12, 13, 17-21). Although there is no consensus in gender studies, most of them report that OM is more frequent among females (1, 5, 6, 12-14, 17, 22, 23), while others indicate a greater prevalence in males (19, 20) or an equal/similar distribution (2, 6, 14, 18, 20). Similarly, the certain predilection of OM to either the maxilla or the mandible is also a matter of controversy. While some studies show that OM occurs almost equally in both jaws (2, 18, 20), a higher incidence in the mandible is reported by most authors (1, 3, 5, 8, 12, 13, 17, 19, 20, 23). The lesion most commonly affects the posterior region in both jaws, and rarely crosses the midline which usually occurs in the mandible (1, 2, 5, 8, 9, 12-14, 18, 20, 23-25). These wide differences regarding gender and location may vary geographically (2).

The majority of OMs are almost always slow-growing and asymptomatic, and are generally revealed by routine radiological examination or due to expansion. However, patients with maxillary/maxillary sinus lesions resulting in neurological disturbance may apply with the complaint

of progressive pain (3, 13, 14, 16, 17). Bone perforation followed by invasion of lesion into the soft tissues can also be observed (1, 13, 14, 18, 26). Pain, paraesthesia, tooth migration and mobility, asymmetry of face, delay in eruption of teeth, speech and mastication disturbances, ill-fitting dentures, and ulceration in oral mucosa are among the other complaints that patients present with (1, 2, 5, 8, 13, 14, 18, 23, 25, 26). Principally, the opinion exists that the duration and severity of the symptoms are consistent with the tumour size (1, 13). Despite its aggressive local growth, OM is believed to never show malignant transformation or cause metastasis (1, 25).

Due to its spreading potential through the maxillary sinus, OM in the maxilla is asserted to be more invasive and destructive in comparison to the mandible by some authors (2, 14, 24). OM in the maxilla may grow unnoticed for a long time, expand quietly into the maxillary sinus, and consequently fill the entire antrum (16, 23). The maxillary tumours may lead to symptoms such as nasal obstruction, exophthalmos, and occasional perforation of the medial and lateral sinus walls due to invasion of the palate, the orbit, and the nasal cavity (3, 14, 17, 22, 23).

Radiographic techniques such as conventional plain radiographs and CT, CBCT, and magnetic resonance imaging (MRI) scans are common are frequently used in comprehensive examination of OM. CT and MRI scans are shown to be more valuable compared to plain radiographs in determining the tumour extention, cortical perforation, soft tissue involvement, and septa pattern exactly (7, 19, 27, 28). On the other hand, conventional plain radiographs may not clearly depict the borders due to the superimposition of many bony structures onto the maxilla, but they can more reliably reveal the displacement and root resorption of teeth (23-25, 28). Besides, CT and CBCT scans are particularly effective in case the lesion is extensive or adjacent to areas where management is challenging (7, 19, 28). CBCT is also very useful in reduction of the patient radiation dose and provides high spatial resolution compared to CT (7). Therefore, it may facilitate the determination of the intraosseous extent of the tumour and it appears convenient to guide the surgeon in the planning of the resection margins.

Radiographically, OMs may display a variable appearance depending on the fibrous tissue and myxoid component amounts, and the cell polarization degree. It may appear as uni – or multilocular radiolucent lesions accompanied by expansion of the cortex, and generally shows a distinctive internal trabecular pattern as “honeycomb”, “soap-bubble”, or “tennis racket” (2, 6, 12, 17, 19, 22-24, 27, 28). The radiological appearances of the tumour are multilocular rather than unilocular radiolucencies (1, 13, 26). The unilocular lesions are usually located in the anterior regions of the jaws, while multilocular lesions are located in the posterior regions (3, 5, 8, 19). As a consequence of internal calcification, a mixed radioopaque-radiolucent appearance of OM has also been reported in 13-20% of cases (1, 19). On the other hand, there are reports of OM cases exhibiting uncommon cortical reaction in the periapical region on conventional radiographs and a “sunray” spicular pattern in CT mimicking osteogenic malignant disease (22, 25, 28). The borders may be well – or poorly-defined, or diffuse (2, 8, 12, 17, 19, 20, 24, 28). Migration of teeth is a rather frequent finding, though resorption of roots appears to be rare (1, 2, 12, 17, 19, 20, 22-24). OM is depicted as scalloping between the roots in tooth-bearing areas (9, 12, 22, 24).

As claimed by Barros *et al*. (15), the radiological characteristics of OMs are determined by the evolution stage of the tumour. Osteoporotic-like areas are produced in early lesions followed by the formation of larger osteolysis areas accompanied by expansion of the cortical bone and infiltration of the adjacent soft tissues. A statistically significant correlation between the lesion size and lesion locularity was demonstrated in the studies conducted by Noffke *et al*. (12), Martínez-Mata *et al*. (13), and Kaffe *et al*. (17), and the larger lesions were more likely multilocular.

The variations in the radiographic manifestation and the overlapping radiographic features with those of other benign and malignant neoplasms make the radiological differential diagnosis of OM difficult (12). A number of lesions should be considered in the radiographic differential diagnosis of multilocular OMs such as ameloblastoma, intraosseous haemangioma, central giant cell granuloma, aneursymal bone cyst, odontogenic keratocyst, fibrous dysplasia, metastatic lesions of the jaws, and periapical, lateral, periodontal and simple bone cysts in unilocular lesions (2, 8, 22, 28). As these facts lead to challenges in making the correct radiological diagnosis, a biopsy is required for an accurate diagnosis (1, 2, 8, 12, 17).

Surgical excision, enucleation, curettage and block resection are the treatment options for OM. Since OM is radioresistant, radiotherapy does not provide any therapeutic benefit (1, 3, 5, 14, 18). Given the size of the lesion, small lesions are recommended to be treated by curettage. On the other hand, complete excision including free margin is suggested for larger lesions in the event that

the tumour is not encapsulated and the myxomatous tissue may infiltrate into the adjacent bone (4, 6). Boffano *et al*. (4) recommended enucleation and curettage as conservative treatment approaches in OM lesions with a diameter less than 3 cm, however larger tumours required segmental resection with subsequent immediate reconstruction. The best possible option to prevent recurrence is radical surgical removal with a 1.5-2 cm margin around the lesion (1).

OM is known to invade bone marrow spaces, and since the cellular neoplastic material can remain in the small bone marrow cavities, it tends to relapse after incomplete removal (12, 21). Regardless of the treatment approach ranging from conservative tumour excision to radical resection depending on the size and behavior of the tumour, a long post-operative follow-up period is recommended, since recurrence typically occurs within a 2-year timeframe following incomplete removal (1, 2, 8, 9, 14-16, 18, 19, 21, 22, 26, 27). However, at least 5 years of follow-up should be recommended as late recurrence cases have been reported (5, 8, 21). Although the recurrence is fairly common with a reported rate ranging from 10% to 33% with an average of 25, complete removal generally provides favourable prognosis in OM cases (1, 2, 6, 8).

In this study, two cases of OM with different clinical and radiological findings are presented. In general, patients' complaints and intraoral presentations were consistent with the literature. Radiological examination revealed well-defined, multilocular hypodense areas with or without hyperdense foci in the CT and CBCT similar to OPTG, but advanced imaging techniques provided better detection of the extension and internal structure of the lesion, the relationship with anatomical structures, and the presence of cortical destruction, expansion or perforation.

Conclusion

CT and CBCT differ from two-dimensional periapical radiography and OPTG because they can demonstrate characteristic radiographic findings and clearly display the tumour in three-dimensional accuracy and perspective. Therefore, the use of advanced imaging is recommended as a useful technique in diagnostic process of the OM in order to assist in imaging the borders and investigating the internal pattern of the tumour.

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