Odontogenic Myxoma of Mandible: A Case Report with Six Months Follow-Up

Mandibulada Odontojenik Miksoma: Altı Aylık Takip ile Bir Olgu Sunumu

Fatih Mehmet COŞKUNSES,[a]
Enver Alper SİNANOĞLU,[a]
Ülkem CİLASUN,[a]
Merva SOLUK TEKKEŞİN[b]

[a]Department of Oral and Maxillofacial Surgery, Kocaeli University Faculty of Dentistry, Kocaeli
[b]Department of Tumour Pathology, İstanbul University Institute of Oncology, İstanbul

Geliş Tarihi/*Received:* 23.10.2014
Kabul Tarihi/*Accepted:* 08.01.2015

Yazışma Adresi/*Correspondence:*
Fatih Mehmet COŞKUNSES
Kocaeli University Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Kocaeli, TÜRKİYE/TURKEY
fcoskunses@gmail.com

ABSTRACT Odontogenic myxoma (OM) is a rare, benign but locally aggressive tumor of the jaws. OMs typically occur between the second and fourth decades and are more common in females than males. According to the World Health Organization (WHO), OM is classified as an odontogenic tumor of ectomesenchymal origin. OMs usually present a asymptomatic swelling and mostly detected incidentally during radiographic examination. They may reach considerable sizes and cause cortical expansion of bone. The treatment choices for OMs are surgical excision by enucleation, curettage, or block resection. Recurrence rates were reported at around 25%, especially with conservative approaches. In this case report, diagnostic, clinical and radiological features of an OM in a 32 years old woman is presented. Surgical treatment, recurrence and follow-up are also discussed.

Key Words: Odontogenic tumors; myxoma; mandibula

ÖZET Odontojenik miksom (OM), çenelerin nadir görülen, ancak lokal olarak agresif bir tümörüdür. Sıklıkla ikinci ve dördüncü dekatlar arasında ve kadınlarda erkeklere oranla daha fazla görülürler. Dünya Sağlık Örgütü (DSÖ) tarafından ektomezenkim kökenli bir odontojenik tümör olarak sınıflandırılır. Asemptomatik olarak şişlik ile karakterize olup sıklıkla radyolojik muayenede tesadüfen tespit edilirler. Etkilenen kemikte kortikal ekspansiyon oluşturur ve ciddi boyutlara erişebilirler. Enükleasyon ile eksize edilmesi, küretaj ya da blok rezeksiyonlar OM'ların tedavi seçenekleri arasında yer alır. Konservatif yaklaşımlarda daha sık olmakla birlikte %25 oranında nüks oranı rapor edilmiştir. Bu olgu raporunda 32 yaşındaki kadın hastada görülen OM'un tanı, klinik ve radyolojik özellikleri ile beraber cerrahi tedavi, nüks ve takip prosedürü sunulmaktadır.

Anahtar Kelimeler: Odontojenik tümörler; miksoma; mandibula

Turkiye Klinikleri J Dental Sci Cases 2015;1(1):12-7

Odontogenic myxoma (OM) is an intraosseous neoplasm characterized by stellate and spindle-shaped cells embedded in an abundant myxoid or mucoid extracellular matrix. When a relatively greater amount of collagen is evident, the term myxofibroma may be used.[1] OM arises from odontogenic ectomesenchyme and resembles the mesenchymal portion of the dental papilla. These develop only in the facial bones and are occasionally related to a tooth that failed to erupt or is missing. In some cases odontogenic epithelium can be detected microscopically.[2]

OMs present an asymptomatic swelling and mostly detected incidentally during radiographic examination. Due to their insidious character,

doi: 10.5336/dentalcase.2014-42160

OMs may reach considerable sizes and cause cortical expansion of bones.[3] It may be difficult to enucleate due to their loose consistency, therefore surgical excision is indicated.[2] In order to ensure that all the neoplastic cells have been removed, it is suggested that, the adjacent "normal-appearing" bone should also be removed with the lesion, considering the difficulties in intraoperative definition of the lesion-normal adjacent bone.[4] The treatment choices for OMs are surgical excision by enucleation, curettage, or block resection. Recurrence rates were reported at around 25%, especially in conservative approaches.[5]

OMs more commonly affect the mandible by a rate of 3:1. In terms of radiographic features, the lesions are usually well defined and often have a corticated margin. However, the outline of some lesions, especially those in the maxilla, is poorly defined. It may also be scalloped between roots of the adjacent teeth. Although most of the time it is described as multilocular, it may be either unilocular or multilocular having a mixed radiolucent-radiopaque internal pattern.[6] There is presence of septa which give OM the multilocular appearance; there may be straight, thin-etched septa (tennis racket like or step ladder pattern) or septa that are curved and coarse with two or three straight septa. The tumor has a tendency to grow along the involved bone without the same amount of expansion seen in other benign tumors.[2]

The present report describes a case of odontogenic myxoma of the mandible with its clinical, radiological and histopathological findings.

CASE REPORT

A 32-year-old woman was referred to Kocaeli University Faculty of Dentistry for investigation of a radiolucent lesion incidentally detected in her mandible during radiographic examination. The patient reported no symptoms and was unaware of the lesion, requiring assessment for prosthetic restoration. In the clinical examination, no signs of swelling or discrepancy from healthy oral mucosa were detected and she had no systemic diseases in her medical history. The panoramic radiograph showed a well-defined, corticated, multilocular radiolucent lesion with poorly defined septa in the non-tooth-bearing right mandibular ramus region (Figure 1). In order to investigate the internal structure, locularity, expansion, and the association with mandibular canal cortex of the lesion, a cone beam computed tomography (CBCT) was taken (Vatech Pax-Flex 3D, Suwon, Korea). Cone beam computed tomography findings revealed radiolucent area with a few strands of delicate or coarse intratumoral trabeculae. Axial sections showed buccal and lingual expansions without cortical destruction (Figure 2). Margins of the lesion were corticated and mandibular canal involvement was detected in the coronal sections (Figure 3).

After evaluation of the radiographic findings, an incisional biopsy was performed and the provisional diagnosis of odontogenic myxoma was made with the presence of spindle shaped cells in myxoid stroma. In the operative intervention, enucleation was performed under general anesthesia and an intact, solid lesion was enucleated surgically with ease. Due to intraoperative surgical impression of the lesion, further resective treatment options were not considered (Figure 4a). The specimen was sent then to the Department of Tumor Pathology at the Institute of Oncology of Istanbul University for histopathologic evaluation (Figure 4b). In gross examination, the specimen revealed grey-white mass with a translucent mucious appearance on the cut surface measuring about 3.5 cm in diameter. Histopathologically, the tumor consisted of stellate, spindle shaped and round cells dispersed in a myxoid stroma (Figure 5a). The stroma contained a few fine collagen fibers. Septum of residual lamellar bone was seen

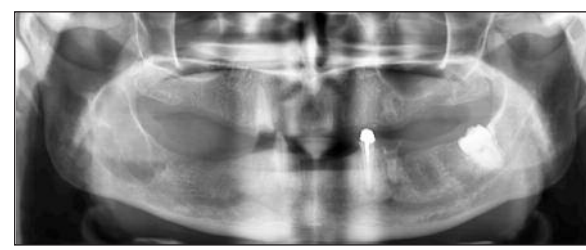

FIGURE 1: Panoramic radiograph displayed a multilocular radiolucent lesion in the right mandibular ramus region.

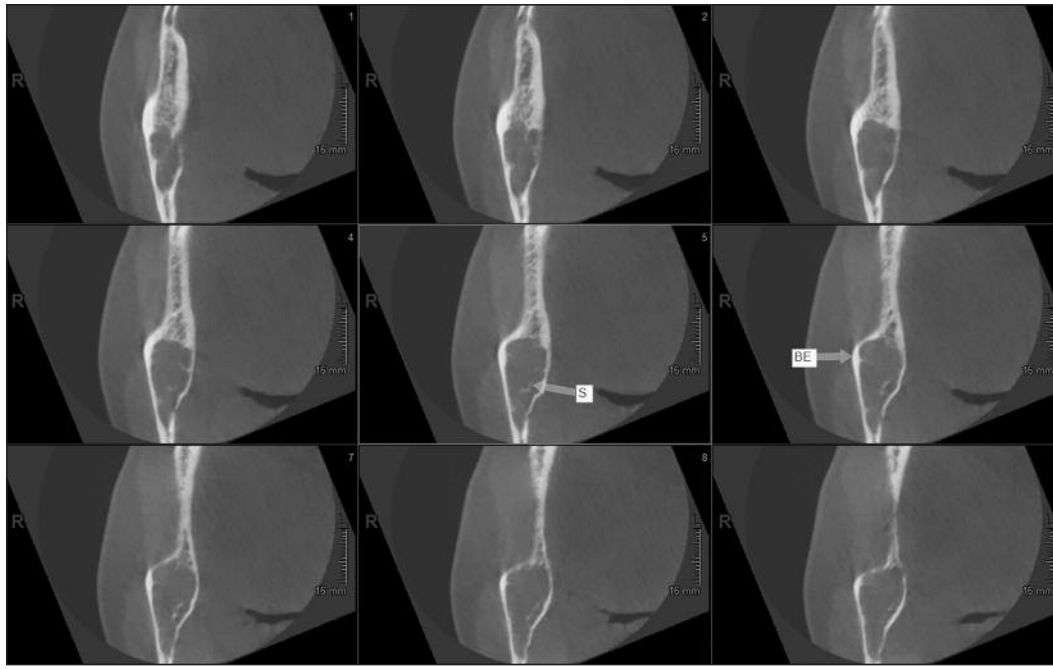

FIGURE 2: Axial view of the lesion at 1 mm slices thickness. Note the expansion of the buccal cortex (BE) and the poorly defined septa (S).

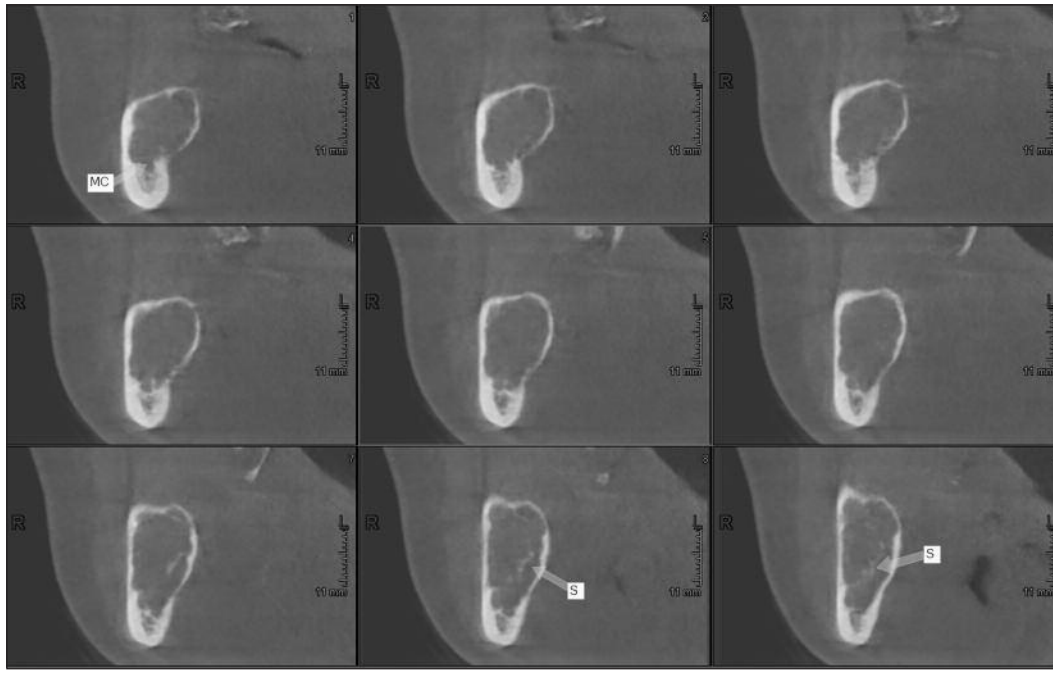

FIGURE 3: Coronal view of the lesion at 1 mm slices thickness. Note the involvement of the mandibular canal (MC) and the poorly defined septa (S).

within the tumor (Figure 5b). The patient is under routine control and in the follow-up of 6 months she was asymptomatic and no evidence of recurrence was found (Figure 6).

DISCUSSION

Since its original description by Thoma and Goldman in 1947, the nature of odontogenic myxomas

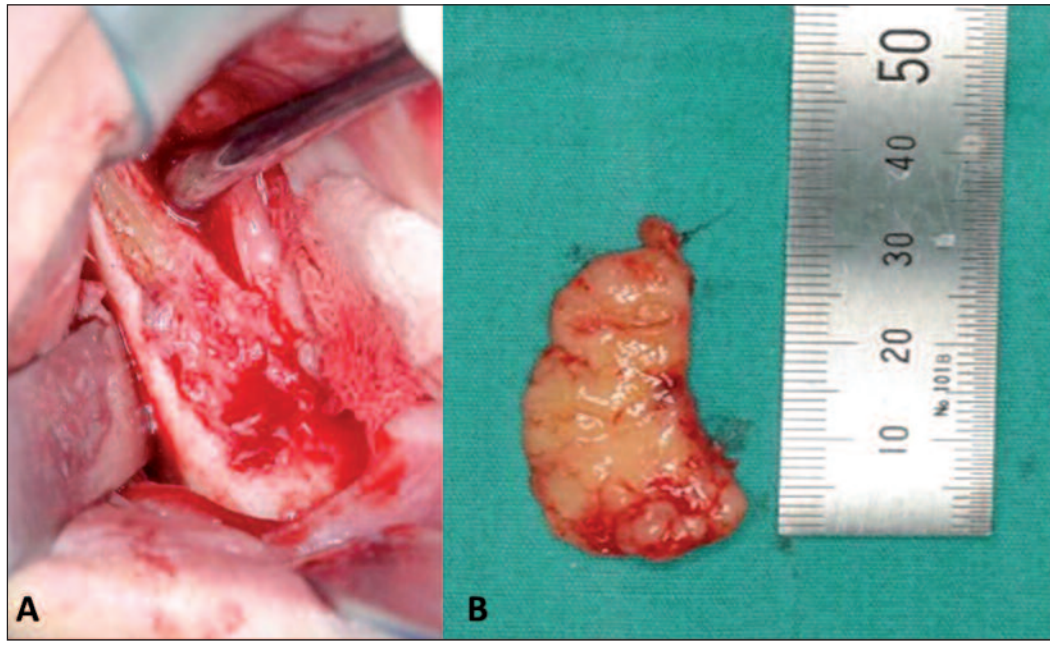

FIGURE 4: (A) Intraoperative view of the lesion. **(B)** Macroscopic view of the lesion.

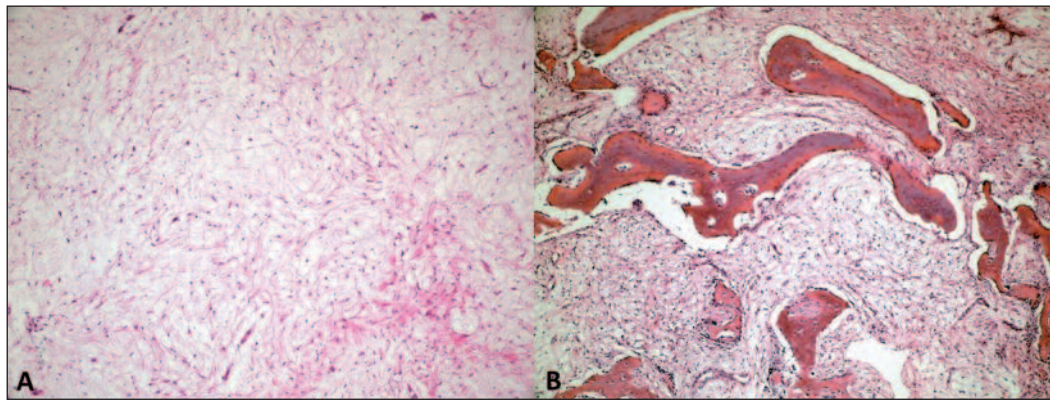

FIGURE 5: (A) Randomly oriented spindle shape cells set in s myxoid background with delicate collagen fibres (H&Ex100) **(B)** Residual lamellar bone inside the tumor (H&Ex100).

has been a matter of controversy.[7] World Health Organization classifies OMs as 'Mesenchyme and/or odontogenic ectomesenchyme with or without odontogenic epithelium'. OMs occur between the second and fourth decades and are more common in females than males (male-to-female ratio 1:1.5).[8] OMs present a slow expansion without symptoms and rarely perforate the cortical bone. It was reported that OMs in the mandible mostly

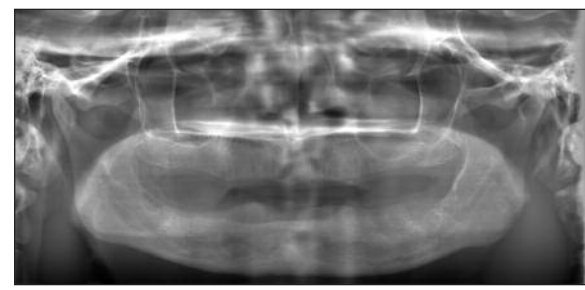

FIGURE 6: Panoramic radiograph of patient 6 months postoperatively.

occur in the premolar and molar areas and rarely in the ramus and condyle as non-tooth-bearing areas.[6] In the presented case, the lesion was located in the ramus region extending to the coronoid process of the mandible. Use of advanced imaging is suggested in diagnostic process of OM in order to show the boundaries and helps the establishment of the internal extent of the tumor and guides in planning the resection margins.[6-9] In the present study, CBCT findings facilitated investigation of the extension of the lesion, the internal structure, the involvement of the mandibular canal, and the presence of cortical destruction.

In 2013 Kheir et al. retrospectively analyzed the radiological images of 33 OM cases. They reported that there was considerable debate and controversy regarding the internal structure and locularity of the tumor. They mentioned about a theory suggesting that the loculation described the stage of tumor development. According to that theory, the tumor was starting out as a multilocular tumor that increased in size together with ongoing resorption of the trabeculae and subsequent conversion to a unilocular format, as an explanation to the combined appearance of honeycomb together with radiolucency with fine trabeculae.[9] On the other hand, based on their findings, they agreed to the postulate that both forms occur independently. Furthermore, they suggested that trabeculae could develop within a unilocular tumor and convert it into a multilocular tumor.[9] In this case report, although the septa were not fine and straight, the lesion was multiloculated as revealed by CBCT.

OM may be mistakenly diagnosed as 'hyperplastic dental follicle' or 'dental papilla of a developing tooth. To avoid the misdiagnosis of these entities, the correlation of clinical and radiological features is essential. Microscopic differential diagnoses also include the other myxoid jaw lesions, such as myxoid neurofibroma, chondromyxoid fibroma, and low-grade myxoid chondrosarcoma. Myxoid neurofibromas include zones that show parallel streaming with organization of collagen and the tumor cells into broad fascicles. The cells also are positive for S100 antibody by immunohistochemistry. Both chondromyxoid fibroma and myxoid chondrosarcoma show at least focal evidence of cartilaginous differentiation and cellular atypia in the latter one.[10,11]

Recurrence of OMs in literature is reported between 25% and 43%. Recurrence of the lesion is adhered to its infiltrative character to the cancellous bone.[7] Due to this entity, some authors recommend surgical resection beyond 0.5 - 1 cm radiographic margins.[12] On the other hand, Batsakis states the main reason for recurrence was thought to be an incomplete removal rather than the intrinsic biological behavior of the tumor.[13] Kansy et al. reported a recurrence fifteen years later after a conservative resective approach.[14] Lo Muzio et al. also reported two cases of recurrences after conservative treatments of OMs.[15] Nevertheless, Lo Muzio et al. recommended that the surgical treatment modality decision should be determined by the size of the lesion. Boffano et al. suggested that conservative treatment by enucleation and curettage should be considered as a treatment choice when the diameter of the lesion is less than 3 cm.[16] Considering the literature, size of the lesion and surgical experience, we decided enucleation and surgical curettage of the lesion in the presented case. In our opinion, conservative surgical approach should be the first treatment choice in such cases that have clear surgical margins. Only after a recurrence, with the presence of close follow-up period of at least 5 years, resective and reconstructive surgical treatments could be performed.

REFERENCES

1. Larheim TA, Westesson PL. Odontogenic myxoma/myxomafibroma. Maxillofacial Imaging, Berlin: Springer-Verlag; 2006. p.58.
2. Karjodkar FR. Benign Tumors of the Jaws. Textbook of Dental and Maxillofacial Radiology, 2nd ed. New Delhi: Jaypee Brothers Medical Publishers; 2009. p.569-70.
3. Manne RK, Kumar VS, Sarath PV, Anumula L, Mundlapudi S, Tanikonda R. Odontogenic Myxoma of the Mandible. Case Reports in Dentistry Volume 2012, Article ID 214704, doi:10.1155/2012/214704.
4. MacDonald D. Radiopacities. Oral and Maxillofacial Radiology-A Diagnostic Approach. 1st ed. Chichester, West Sussex; Ames, Iowa: Wiley-Blackwell; 2011. p.122-3.
5. Rocha AC, Gaujac C, Ceccheti MM, Amato-Filho G, Machado GG. Treatment of recurrent mandibular myxoma by curettage and cryotherapy after thirty years. Clinics (Sao Paulo) 2009;64(2):149-52.
6. White SC, Pharoah MJ. Oral Radiology: Principles and Interpretation. 6th ed. St. Louis: Mosby; 2008. p.386-7.
7. Miranda Rius J, Nadal A, Lahor E, Mtui B, Brunet L. Unusual presentation of localized gingival enlargement associated with a slow-growing odontogenic myxoma. Int J Oral Sci 2013;5(3):172-5.
8. Kaffe I, Naor H, Buchner A. Clinical and radiological features of odontogenic myxoma of the jaws. Dentomaxillofac Radiol 1997;26(5): 299-303.
9. Kheir E, Stephen L, Nortje C, van Rensburg LJ, Titinchi F. The imaging characteristics of odontogenic myxoma and a comparison of three different imaging modalities. Oral Surg Oral Med Oral Pathol Oral Radiol 2013; 116(4):492-502.
10. Neville BW, Damm DD, Allen CM, Bouquot JE. Odontogenic cysts and tumors. Oral and Maxillofacial Pathology. 3rd ed. Missouri: Saunders Elsevier; 2009. p.729-31.
11. Manjunath S, Gupta A, Swetha P, Moon Nj, Singh S, Singh A. Report of a rare case of an odontogenic myxoma of the maxilla and review of literature. Ann Med Health Sci Res 2014;4(Suppl 1):S45-8.
12. Leiser Y, Abu-El-Naaj I, Peled M. Odontogenic myxoma--a case series and review of the surgical management. J Craniomaxillofac Surg 2009;37(4):206-9.
13. Batsakis JG. Myxomas of soft tissues and the facial skeleton. Ann Otol Rhinol Laryngol 1987; 96(5):618-9.
14. Kansy K, Juergens P, Krol Z, Paulussen M, Baumhoer D, Bruder E, et al. Odontogenic myxoma: diagnostic and therapeutic challenges in paediatric and adult patients--a case series and review of the literature. J Craniomaxillofac Surg 2012;40(3):271-6.
15. Lo Muzio L, Nocini P, Favia G, Procaccini M, Mignogna MD. Odontogenic myxoma of the jaws: a clinical, radiologic, immunohistochemical, and ultrastructural study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1996; 82(4):426-33.
16. Boffano P, Gallesio C, Barreca A, Bianchi FA, Garzino-Demo P, Roccia F. Surgical treatment of odontogenic myxoma. J Craniofac Surg 2011;22(3):982-7.