A Giant Keratocystic Odontogenic Tumour: Surgical Treatment and Unusual Reconstruction with Autologous Fat Graft: Surgical Technique

Dev Keratokistik Odontojenik Tümör: Cerrahi Tedavi ve Otojen Yağ Dokusu Grefti ile Rekonstrüksiyon

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ABSTRACT Keratocystic odontogenic tumour (KCOT) is a common developmental benign odontogenic neoplasm in the jaws. In this paper, a case of a large KCOT and unusual reconstruction with autologous fat graft are reported. A 73-year-old woman was referred to our clinic with a complaint of right preauricular swelling and pain. Radiographic examination showed large cystic lesion that involved right side of the mandibular ramus and extend from the right mandibular molar area to the subcondylar region. Considering the general status of the patient and the size of the defect an autologous fat graft was planned. The large bone defect was filled with autologous fat graft from the abdomen. Autologous fat transfer may be an alternative technique for reconstruction of the large intra oral defect in selected cases in which other reconstruction methods are not feasible because of the general status of the patient and the size of the lesion .

Key Words: Clinical protocols, odontogenic cysts

ÖZET Keratokistik odontojenik tümör (KCOT) çenelerin sık görülen gelişimsel odontojenik benign neoplazmıdır. Bu yazıda, büyük bir KCOT olgusu ve otojen yağ dokusu ile sık olmayan rekonstrüksiyonu rapor edildi. Yetmiş üç yaşında kadın hasta kliniğimize sağ preauriküler şişlik ve ağrı şikâyeti ile başvurdu. Radyografik muayene, sağ mandibular ramusu içine alan, sağ mandibular molar bölgeden subkondiler bölgeye uzanan büyük kistik lezyon gösterdi. Olgumuzda büyük kemik defekti karından alınan otojen yağ dokusu grefti ile dolduruldu. Otojen yağ dokusu transferi, hastanın genel durum bozukluğu ve defektin büyüklüğü gibi faktörler nedeniyle diğer rekonstrüksiyon yöntemlerinin uygulanamadığı, seçilmiş olgularda büyük intraoral defektlerin rekonstrüksiyonunda alternatif bir teknik olabilir.

Anahtar Kelimeler: Tedavi yöntemleri, odontojenik kist

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he nature of the odontogenic keratocyst (OKC), also known as KCOT is controversial. The aggressive behaviour and high recurrence rate of OKC suggests a true neoplastic potential and has prompted the World Health Organization Working Group to classify the OKC as a benign tumour. It is locally aggressive and has a higher rate of recurrence than other odontogenic cysts. They usually develop between the ages 10-40 years. A slight male predilection is usually seen. KCOT grows slowly and causes no symptoms until a swelling becomes noticeable. Around 60-80% of the reported cases occurred in the posterior body of the mandible. Its typical radiographic presentation is that of a well-defined, unilocular or mul-

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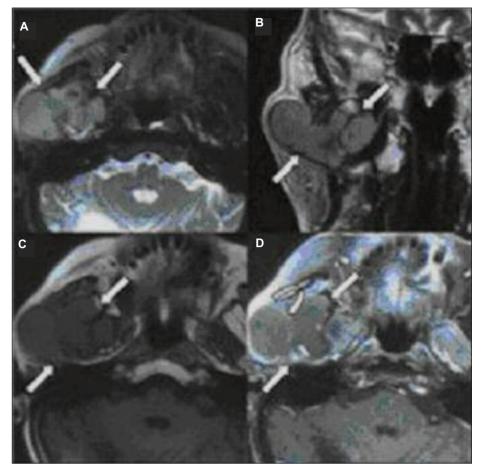


FIGURE 1: Preoperative MRI are shown. A. Transverse fat saturated T2 weighted section. Arrows indicate the slightly hyperintense multiseptate lesion with destruction of the right mandibular condyle and ramus. B. Coronal T2 weighted section. The lesion seems to be less intense when compared with fat saturated sequence. C. Transverse T1 weighted section. The lesion is seen as hypointense. D. Transverse fat saturated T1 weighted section after contrast medium administrated. Note that the tumour wall and septa are enhanced (curved arrow).

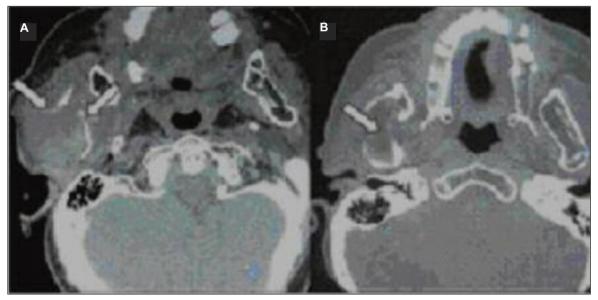


FIGURE 2: Preoperative (A) and postoperative (B) CT images. The lesion is indicated by arrows at the right side. The destruction of the bone is evident. Arrow in the Panel B indicates the graft material of fat density filling up the surgical cavity.

Ear-Nose-Throat Diseases Çakır Özkan et al

tilocular radiolucency and expansion of the mandibular buccal and medial cortical plate.^{2,3}

Two variants of this lesion are well known; the sporadic cyst and the cyst associated with the nevoid basal cell carcinoma syndrome. Both variants of the KCOT are believed to originate from remnants of the dental lamina. The lesion may be effectively treated with enucleation and curettage. Some authors have suggested the resection of KCOT with 5 mm linear margins.^{2,3}

The following describes a case of KCOT in the right mandibular posterior body and ramus that was treated by excision and was reconstructed with an autologous fat transfer.

A 73-year-old woman was referred to our clinic with right preauricular swelling and pain. Clinically, the right preauricular region and ramus of the mandible was swollen and it was tender on palpation. The patient had been operated on this region 13 years ago. Computerized tomography and magnetic resonance imaging (MRI) of the mandible showed a large lesion that involved the right side of the mandibular ramus and extended from the right mandibular molar area to the subcondylar region (Figure 1A, B, C, D and Figure 2A). Based on clinical, radiographical and fine needle aspiration biopsy results, it was diagnosed as KCOT.

The operation was performed extraorally using modified blair incision under general anaesthesia. The lesion expanded the medial and lateral cortical plates and reached medially to the pterygopalatin fossa, but medial cortical plates were intact. Enucleation and curettage were performed. Considering the general status of the patient and the size of the defect an autologous fat graft was planned to fill the large defect. The cavity was obliterated with subdermally dissected fat graft from the abdomen. The postoperative period was uneventful. The histopathologic analysis of the specimen confirmed the diagnosis of the KCOT (Figure 3A, B). The patient was followed up for eight months and no local recurrence was noticed (Figure 2B).

DISCUSSION

KCOT are slowly growing, keratinizing, epithelial-lined tumours. The aggressive behaviour and

high recurrence rate of KCOT suggests a true neoplastic potential. The authors suggest that genetic alterations support the neoplastic nature of KCOT. Although additional molecular investigations, including epigenetic studies, should be performed, the current scientific evidence supports the neoplastic nature of this lesion.^{1,4,5}

Similarly, our patient had undergone cyst enucleation for right mandibular region 13 year ago. Therefore, we believed that this tumour was a recurrent lesion in keeping with the literature.

There is no agreement on the treatment of KCOT. Enucleation following marsupilisation or only enucleation and the resection of KCOT with linear margins can be performed. Most of the sporadic KCOT's may be effectively managed with a thorough enucleation and curettage surgery. MacIntosh suggested the resection of KCOT with 5 mm linear margins as the preferred primary method of treatment and reported 37 patients with 43 lesions emphasizing the efficacy and superior results of resection over all other treatment methods.³

A number of surgical procedures have been suggested for the reconstruction of defects in the jaws after ablative surgery including autografts, allografts, primary closure, regional flap and distant flaps. The choice of the reconstruction is based on the type and size of the defect.^{6,7} Autogenous bone grafts were shown to be superior to allogeneic bo-

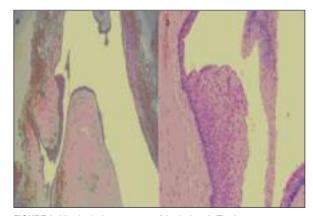


FIGURE 3: Histological appearance of the lesion. A. The low power appearance of the tumor wall (HE, x5 magnification). B. Close appearance of cyst wall and mature lining squamous epitel. Distinctly thickened squamous epitel has continuity with mature squamous epitel which is a thinner layer (HE, x30 magnification).

Çakır Özkan ve ark. Kulak-Burun-Boğaz Hastalıkları

ne and alloplasts in terms of function, form and adaptability. However, there are limitations such as the inadequate supply and donor-site morbidity. Allografts have been used in reconstructive procedures, but their volume and lack of osteocompetent cells limit their use.⁶

Autologous fat transfer techniques for facial reconstruction and maxillofacial surgery have also found a place in the contemporary oral and maxillofacial surgery practice. Buccal fat pad flap has been used in reconstruction of small defects in the oral cavity. The indications for use of the buccal fat pad include defects excision of a benign or malignant tumour preferably smaller than 5 cm. The complications associated with the transfer of autologous fat are limited such as misplacement of the graft, infection and loss of graft volume. The autologous fat graft serves as a bed for secondary granulation by reducing dehiscence in the soft tissue

layer and it physically aids in closure by obliterating the defect. 8.9 In our case, considering the general status of the patient and the size of the defect, an autologous fat graft from the abdomen was used to fill the large defect. Healing was perfect with no complication after the defect was reconstructed with an autologous fat graft.

MRI can be used to distinguish local recurrence and fat graft in the postoperative follow-up period. The patient's progress was monitored monthly for the first six months following surgery and continues to be examined yearly.

It is concluded that autologous fat transfer may be an alternative technique for reconstruction of the large intra oral defect in selected cases in which other reconstruction methods are not feasible because of the general status of the patient and the size of the lesion.

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