CASE REPORT

Treatment of mandibular odontogenic keratocyst by decompression with a customized removable device: A case report and literature review*

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ABSTRACT

Treatment of mandibular odontogenic keratocyst by decompression with a customized removable device: A case report and literature review

Odontogenic keratocyst (OKC) is a benign, rare intraosseous tumor, but in some cases it may be aggressive and infiltrate into neighboring tissues. These cysts have a high recurrence rate and show specific histopathological features. Decompression is an established technique for the treatment of many large odontogenic cysts ranging from small and simple cysts to odontogenic keratocysts and has recently become a popular alternative technique. However, this approach requires that the opening in the cyst cavity does not close. There are several techniques described in the literature for OKC treatment with decompression. Here, we present the results from the treatment of a 17-year-old female patient with high-volume OKC associated with an impacted wisdom tooth in the right posterior ramus region, by using a customized decompression plate.

KEYWORDS

Customized removable device, decompression, odontogenic keratocyst

ÖZ

Mandibular odontojenik keratokistin kişiye özel çıkarılabilir bir aparey yardımıyla dekompresyon tedavisi: Vaka sunumu ve literatür derlemesi

Odontojenik keratokist (OKC) iyi huylu, nadir görülen intraosseöz bir tümör olmasına rağmen bazı durumlarda agresif yapıda olabilmekte ve çevre komşu dokulara infiltrasyon gösterebilmektedir. Bu kistler yüksek bir nüks oranına sahiptirler ve spesifik histopatolojik özellikler gösterirler. Dekompresyon, basit-küçük kistlerden odontojenik keratokistlere kadar birçok büyük odontojenik kistin tedavisi için kabul görmüş bir tekniktir ve son zamanlarda popüler bir alternatif teknik haline gelmiştir. Fakat bu yaklaşım kist kavitesinde açılan boşluğun kapanmamasını gerektirir. Literatürde bu amaç için tanımlanmış çeşitli teknikler bulunmaktadır. Bu bilgiler ışığında bu sunumda sağ posterior ramus bölgesinde gömülü yirmi yaş dişi ile ilişkili büyük hacimli OKC bulunan 17 yaşındaki bayan hastanın özelleştirilmiş dekompresyon plağı ile tedavisini mevcut literatür eşliğinde sunmaktayız.

ANAHTAR KELİMELER

Kişisel çıkarılabilir aparey, dekompresyon, odontojenik keratokist

Odontogenic keratocyst (OKC), first described by Phillipsen in 1956, is a developmental cyst that originates from dental lamina.[1] OKC is usually observed in the mandibular ramus and the angulus regions and radiologically display a round or oval shaped unilocular or multilocular structure. It is usually seen in males in the second and fourth decades of life.[2] Since OKCs are often associated with impacted teeth, differential diagnosis should be applied in order to differentiate from other pathological lesions.[3] These cysts have been one of the most debated lesions in the literature due to their histopathological features, high recurrence rate and aggressive character.[4-6] In 2005, OKC was renamed as "keratocystic odontogenic tumor" by the World Health Organization (WHO) because of its neoplastic properties.[7] However, it was reincluded in the cyst classification by WHO in 2017.[2]

In the past, OKCs were believed to be highly aggressive forms of odontogenic cysts, and therefore more aggressive treatments such as curettage with peripheral ostectomy, curettage plus application of Carnoy's solution, cryotherapy, localized en bloc resection or mandibular segmental resection were recommended for the treatment instead of simple enucleation.[5,8-11] However, since OKCs can have very large dimensions, aggressive treatments can cause damage to adjacent tissues and anatomical structures (such as maxillary sinus, alveolar inferior and mental nerve) especially in young and elderly patients. Therefore, decompression and marsupialization methods, which are defined as conservative treatment options, have emerged as important alternative treatment methods. In the literature, both of these methods were shown to be effective for the treatment of OKCs.[12,13] Enislidis *et al*[14] treated 20 large-volume mandibular cysts with decompression and recommended decompression as an effective treatment option, despite the length of

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the treatment duration. Anavi *et al*[15] used decompression for 73 odontogenic cysts and performed long-term follow-up of the patients, and they suggested decompression as the primary treatment option for odontogenic cysts while they also strongly recommended secondary surgery after decompression for the treatment of aggressive cystic lesions. Maurette *et al*[16] have treated 30 OKC cases with decompression, and reported that this approach provides a valid treatment option due to low morbidity and similar recurrence rate to other methods in the literature, while also suggesting longer follow-up. Pogrel *et al*[17,18] have suggested that decompression and marsupialization should be preferred as treatment options for OKCs with no recurrence in their study. For this reason, these two methods became popular conservative treatment options in the treatment of large cystic lesions and were widely used compared to others. In this context, we also preferred decompression therapy in order to prevent possible tissue loss and tissue damage in a young patient in the present study.

Although decompression and marsupialization are both recommended as treatment options, the terms are technically different, because marsupialization consists of the suturing of the pathological tissue with the healthy mucosa, forming a pouch, whereas decompression uses a device to maintain the patency of the cyst and requires its daily irrigation.[12,13,17] Thus, it reduces the size of the cyst by altering its osmotic pressure and favors the formation of new bone tissue. In the literature, various modified decompression devices such as cylindrical polyethylene drainage or infusion tubes, or plastic stents have been used for this purpose. The purpose of this study was to present a new easy to use decompression device which can reduce the trauma to soft tissues after fixation and loosening of the polyethylene tubes to the cavity and to eliminate the tube fixation problem.

CASE REPORT

A 17-year-old female patient applied to our clinic for routine controls. In the intraoral examination, oral and dental tissues were normal and no pathology was found. In the extraoral examination, no cervical lymphadenopathy and no asymmetry was seen. After taking medical history and physical examination, the patient was sent for radiological examination. The panoramic film showed an asymptomatic lesion associated with the impacted tooth in the ramus region (Figure 1). After the patient and her parents have been informed and their consent was obtained, decompression therapy was planned considering the age of the patient and the size of the lesion. Under local anesthesia, a bone window was opened in the lesion area. The contents of the lesion were removed and biopsy was taken from cyst epithelium. Based on the clinical and radiological findings of the patient, our differential diagnosis included OKC, radicular cyst and dentigerous cyst of the mandible. After the histopathological evaluation, OKC was diagnosed. During the operation, cyst cavity was filled with iodoform gas and then an impression was taken from mandible. Decompression device was obtained on the plaster model (Figure 2). Prepared appliance was positioned in the mouth and the patient was informed about how to use it. She was instructed to irrigate the cavity with a 0.12% chlorhexidine solution using a plastic syringe. On the 3rd and 6th months of the patient's examination, ossification foci began to appear on the radiograph and with the reduction of the lesion, it was seen that the impacted tooth continued to grow. In the 9th month, the impacted tooth was removed with a second operation. After 18 months, the intraoral healing was normal and there was an ordinary contrast image on the tomography and normal ossification was detected in all over the lesion (Figure 3). The control visits have been performed and are also planned for the future follow-up in case of the possibility of recurrence.

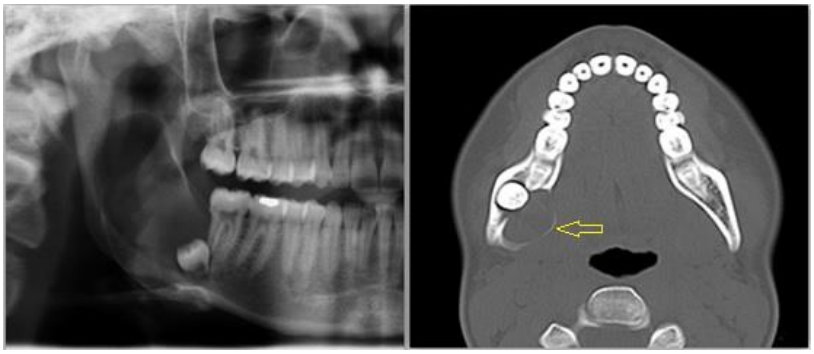

Figure 1.

Radiographic and CT view of the patient's mandible before operation

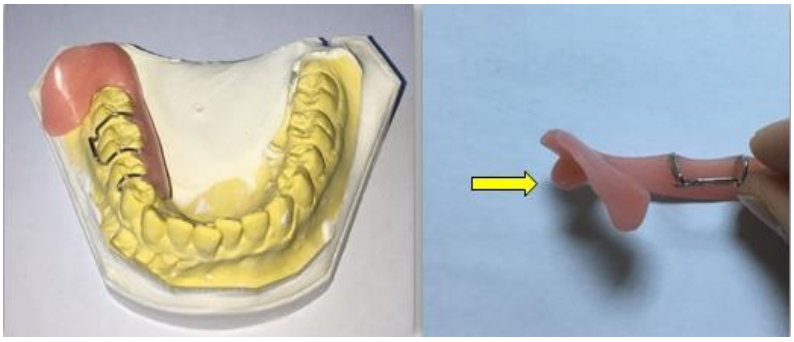

Figure 2.

Personalized decompression device on the model

Figure 3.

Post-operative healing on 18th month new ossification areas are seen

DISCUSSION

It is known that the cysts grow due to resorption following the increased osmotic pressure in the epithelium, and prostaglandins and growth factors play important roles in the formation of this process.[19] Especially in large-sized lesions, decompression therapy removes the cyst fluid, and the decompression device ensures continuous washing of the cyst cavity. It is thought that the factors that are effective in the inflammatory processes are also removed from the area through this procedure. For example, Interleukin-1 has been shown to be an inflammatory cytokine that plays an important role in the growth of OKCs. Immunohistochemical studies showed that the high levels of interleukin-1a in OKC were significantly reduced after decompression treatment.[20]

Several studies have shown that a number of changes occur in the cyst epithelium when keratocysts are opened into the oral cavity. Researchers observed that the cyst epithelium thickens after decompression or marsupialization; and epithelium starts to resemble normal oral mucosa over time both histologically and immunohistologically.[17,21] Compared to other treatment options, both decompression and marsupialization have distinct advantages, and therefore, their usage has been becoming more prevalent.

Critics of the use of marsupialization or decompression for the treatment of OKC argue that this technique does not allow complete removal of cysts and, therefore, epithelial proliferation continues which can increase the likelihood of the recurrence of the symptoms.[9] However, other researchers stated that they observed no recurrences in their studies.[12, 17]

Another important benefit of decompression therapy is fewer side effects compared to radical surgery, including large-scale tissue loss, pathological fracture formation, nerve and adjacent anatomic tissue damage.[12,13] Researchers have argued use of marsupialization or decompression instead of radical surgery to reduce the volume of the lesion, allowing curettage or enucleation to be done more easily and without risk.[12,22]

In the literature, several decompression devices (tubes or stents), which were produced from synthetic polymers such as various plastics or polyethylene, were successfully used to prevent closure of the opening in the cyst cavity during decompression or marsupialization treatments.[14,17,22-28] These devices are generally cylindrical or in the shape of a surgical drain. The appropriate technique and material to be used for this treatment should be selected depending on the condition of the patient, and the size, location and type of the pathology. While some researchers have used intravenous infusion tubes, intubation tube or nasopharyngeal tube as a decompression device,[22,28] others have used a wide mouth drain made by using a luer syringe.[26] Tolstunov[25] reported that intravenous tubing, nasal cannula and urethral catheter could be used for the treatment of odontogenic cysts. These devices are not only widely available and inexpensive, but also their position in the cavity can be monitored during treatment due to their radiopacity. Almost all of the aforementioned devices are fixed to the mucosa or the bone in various ways (suture, wire, etc.) inside the oral cavity,

however, this causes the section of the device that stays in the cyst cavity remain free at the apical. We think that this can cause a problem during treatment because it is inevitable that the tube gets loose in a constantly moving environment. Several studies have also pointed out that the sutures might be lost, re-suturing might be necessary, stents might be dislocated or buried, or soft tissue might be buried or traumatized as a result of this method.[22,25,26,28] Since decompression therapy requires a lengthy duration, these problems might present significant disadvantages. For this reason, we preferred to use a customized acrylic decompression plate personalized by using a model created by using measurements from the cyst cavity of the patient.

Various decompression tubes in different styles and designs have been used in the literature and each design was recommended by respective authors. Swantek *et al*[24] have designed a decompression stent consisting of two plastic pieces nested with one wide, one narrow tip. While the lower part of the stent was retained in the cyst cavity, the wide part of the stent was fixed to the bone adjacent to the opening of the cavity by using mini screws. They reported that in this way the device remained attached during the long-term treatment period and the patient could easily wash the cyst contents daily. However, we think that this technique is expensive and is not suitable for all patients.

Some researchers have used wires to immobilize the decompression tube. Kolkythas and colleagues passed wires through the decompression tubes and fixated them to the adjacent tooth cervical by winding in a case study on 22 patients.[23] The authors stated that this technique is well tolerated but care should be taken not to damage the mucosa and gingiva. Costa *et al*[27] improved this technique and inserted an orthodontic stainless-steel wire with a ring on one end through the tube which is in the cyst cavity and secured it to the adjacent tooth by using a composite. The authors stated that this method is useful but can only be applied when there is a tooth adjacent to the cyst cavity. In another study, Zhu *et al*[22] connected the wire that passed through the decompression tube to the brackets placed on the adjacent tooth. They reported that with this technique, the wires can easily be removed, the length and position of the tube can be adjusted and it can be easily cleaned. However, since these techniques can only be applied in the presence of adjacent teeth, they cannot be used in toothless jaws, the tube lumen can be constricted because of the wire, it can create food retention and thus cause problems in the mucosa or gum. Therefore, in the present study, an acrylic plate that was personalized for the patient's mouth and the cyst cavity was prepared, and the opening of the cyst cavity was kept open during the treatment, while no trauma or secondary infection was observed in the neighboring tissues. Carter *et al* have similarly designed and used an individualized device by taking measurements from the cyst cavity and recommended its usage for OKC treatment.[29] On the other hand, there are some circumstances that limit the use of the decompression plate that we have designed. Examples are cases in which patient co-operation cannot be established, and the patient insists on the use of a different type of treatment. Another important issue is properly informing the patient. The patient should be taught in detail how to remove the plaque and how the cyst cavity should be irrigated with a syringe.

It should be kept in mind that decompression therapy is a lengthy process for both the patient and the physician, thus patient follow-up is very important and care should be taken in choosing the appropriate decompression device personalized for the patient and patient's lifestyle in order to provide the optimal treatment.

CONCLUSION

Overall, we recommend the use of customized acrylic plaques for decompression and / or marsupialization treatment for cases with large volume cysts (odontogenic keratocysts, radicular cysts, dentigerous cysts, etc.) in the jaws. We believe that this design is useful because it does not require fixation procedures such as using sutures, wire or screws, it has low cost, can be easily applied, and is an effective treatment method.

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