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A Congenital Sublingual Teratoid Cyst Extending to the Hyoid Bone: A Case Report

Hiyoid Kemik Seviyesine Uzanan Konjenital Sublingual Teratoid Kist Vakası

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ABSTRACT

Dermoid cysts are cavities which are lined by epithelium containing skin appendages. They are classified into three groups as dermoid, epidermoid and teratoid according to the histological type of tissues lining the cyst wall. The most effective method for the treatment is the surgical excision. This case reports a giant teratoid cyst located in the floor of the mouth descending to the hyoid bone level via transgeniohyoid route, which is surgically removed by intraoral approach. The cyst was excised via intraoral approach after decompression . The pathological report was a teratoid cyst.

Keywords
Congenital; excisionase; decompression

ÖZET

Dermoid kistler, içerisinde cilt eklerini barındıran epitel ile çevrili kavitelerdir. Kist duvarını oluşturan dokunun histolojik tipine göre dermoid, epidermoid ve teratoid olarak üç grupta sınıflandırılır. Bu kistlerin tedavisinde en etkili metod, cerrahi eksizyondur. Sunulan bu vaka, intraoral yolla cerrahi eksizyon uygulanan ağız tabanı yerleşimli hiyoid düzeyine kadar uzanan büyük bir teratoid kist olgusudur. Kist dekompresyon sonrasında intraoral yaklaşımla cerrahi eksizyon yapılarak tedavi edildi. Patoloji sonucu teratoid kist olarak rapor edildi.

Anahtar Sözcükler
Konjenital; eksisionaz; dekompresyon

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INTRODUCTION

Dermoid cysts are cavities which are lined by epithelium containing skin appendages (such as hair follicles and sebaceous glands). Of all dermoid cysts, 2-7% affect the head and neck region and they often develop at the ages of 15 to 25 years.[1,2] Within this region, dermoid cysts are most commonly located in the lateral third of the eyebrow. Dermoid cysts are histologically classified as dermoid, epidermoid, and teratoid cysts. Teratoid cysts, which account for approximately 1.8% of all dermoids, are extremely rare.[3] The lining of teratoid cysts consists of not only simple squamous or respiratory epitelium, but also tissues generated from ectoderm, mesoderm, and/or endoderm (e.g., hair and teeth).[4] These cysts arise from entrapment of the germinal epithelium during closure of the mandibular and hyoid branchial arches. These tumors usually appear as a painless, slowly growing, and nontender mass located in the sublingual, submental, or submandibular region. Teratoid cysts of the floor of the mouth, which occur rarely in infants, are generally diagnosed in adults at the second and third decades of life.[3,5]

The most effective treatment for dermoid cysts is surgical excision, which can be intraoral, extraoral, or a combination of both depending on the location and the size of the cyst. This study reports a case of a twenty-four year old male who presented with a giant teratoid cyst of the floor of the mouth extending to the hyoid bone through geniohyoid muscles. We discuss the clinical steps taken to achieve an accurate diagnosis and treatment of the cyst via a solely intraoral approach without any external incision.

CASE REPORT

A twenty-four year old male referred to our clinic with the complaint of a fast growing, tender sublingual swelling for about three weeks. The patient experienced difficulty in swallowing and breathing. Clinical examination revealed a well-defined, firm mass with a smooth surface in the floor of the mouth. This mobile mass, measured approximately 4.0 cm x 4.0 cm and was covered with apparently normal mucosa. On examination, a soft swelling was palpated in the submental region. A computerized tomography (CT) of the neck showed a 35 x 45 mm well-defined mass in the submental region reaching down to hyoid cartilage level (Figures 1, 2).

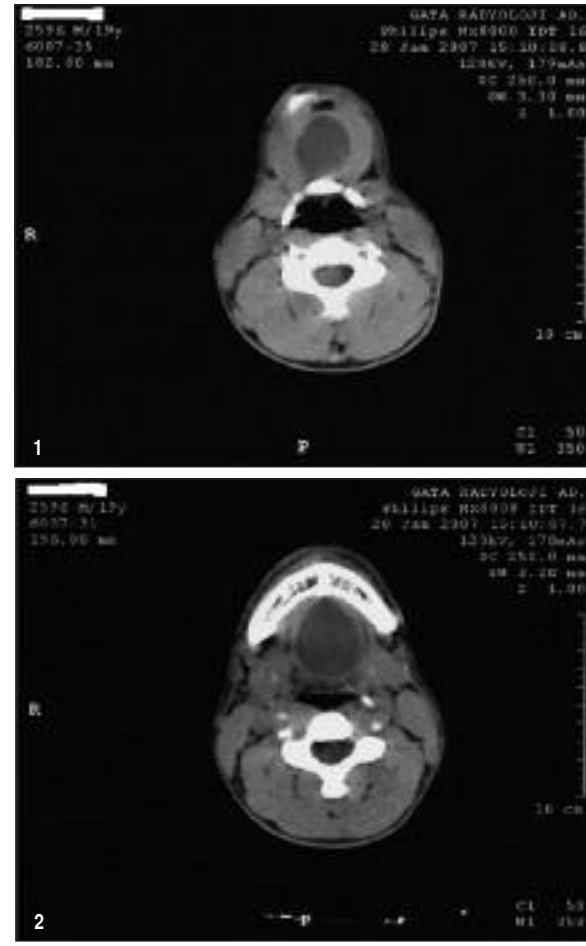

Figure 1, 2. Computerized tomography of the teratoid cyst involving submental and pre-hyoid area.

The wall of the mass was 3 mm in thickness and it was compatible with thyroglossal cyst. The ultrasonographic and color Doppler examination revealed that the mass has no circulatory activity and that it presents a heterogeneous hypoechoic structure. In addition, fine needle aspiration biopsy showed non-specific inflammation. On the basis of these findings, total excision of the cyst was performed in order to obtain a certain diagnosis and to relieve the symptoms of the patient. Intraoral approach was preferred to achieve complete excision with minimal morbidity and scarring.

After applying a retraction suture to the tip of the tongue, a superficial incision was made over the mucosa covering the cyst vertically in the medial region of the ventral surface of the tongue and floor of the mouth. The anterior extension of the incision was performed between the Wharton papillae, taking care not to injure the ducts. The dissection was carried out in the submucosal plane with care not to traumatize the cyst wall or the

mucosa. Approximately 25 cc of yellowish, blurry fluid was aspirated in order to facilitate adequate exposure of the deep side of the cyst and to ease the surgical manipulations (Figure 3, 4). The cyst was descending between the geniohyoid muscles and reaching down to the level of anterior border of the hyoid bone. Eventually the cyst was totally excised and submitted for pathological examination.The mucosal incisions were closed with 4.0 vicryl sutures, special attention was given not to damage the ductus and the Wharton papillae (Figure 5). No drain was placed. The patient was kept intubated over night and in order to avoid air way obstruction due to tongue edema and/or hematoma intravenous steroid regimen was ordered. Postoperative period was uneventful and the saliva drainage through the wharton papillae was not impaired .

Figure 5. The macroscopic view of cyst after total excision.

Figure 3, 4. Perioperative images of surgical intervention.

The pathological examination of the specimen confirmed the presence of a cyst, lined by squamous epithelium, macroscopically containing hair inside. In microscopic examination inflamatory reaction was significant in the epithelium and hair particles were phagocyted by macrocytes within the cyst wall. Thus the diagnosis was reported as mature cystic teratoma.

DISCUSSION

Dermoid cysts of the floor of the mouth vary in size with the largest reported cyst being 10.0 cm x 8.0 cm,[6,7] and as the cyst grows larger, it conceivably impedes breathing and swallowing. Although the teratoid cyst in the present case was relatively smaller in size, the symptoms of the patient progressed rapidly. During the preoperative period, a significant increase in the size of the cyst was observed day by day. The rapid enhancement could be explained by the presence of the infection in the cyst, which was confirmed by the pathologic examination.

Moreover, the swelling (due to the cyst) at the base of the tongue caused difficulty in endotracheal intubation. Surgical excision of the dermoid cysts of the floor of the mouth can be performed via an intraoral, extraoral, or a combined intraoral and extraoral approach. Although a common opinion in the past is to choose the surgical access according to the size of the cyst, the size of the lesion is not an adequate factor in itself since the size of the jaws of patients (newborns to adults) in

which a dermoid cysts may occur varies.[6] The location of the lesion is also a determining factor in choosing the surgical approach.

The dermoid cysts can be classified into three classes according to the location of the cyst relative to the geniohyoid muscle: 1) cysts that are located above the geniohyoid muscle, 2) cysts that are located below the geniohyoid muscle, and 3) transgeniohyoid cysts that pierce through the geniohyoid muscle barrier. The oral approach is the treatment of choice when the cyst is above the geniohyoid muscle, whereas the extra-oral approach is mandatory when the cyst lies under the geniohyoid muscle.[8]

For transgeniohyoid cysts, two different approaches have been reported in the literature: an oral route with cyst decompression, and a combined oral and dermal route. Di Francesco et al.[9] and Zeltser et al.[10] recommended decompression and removal of large cysts via the oral route, where the decompression of the cyst partially removes its contents and makes the dissection of the cyst easier. On the other hand, the combined approach, which enables adequate visualization, was recommended if proper care must be taken not to harm important adjacent structures.[6]

The teratoid cyst in the present case was a transgeniohyoid cyst reaching to the level of the hyoid bone. Complete excision of this lesion through solely intraoral access was technically challenging especially while dissecting the infero-medial surface of the cyst. However, aspiration of the cyst content before its extirpation enhanced the exposure and enabled the total excision of the cyst.

CONCLUSION

In conclusion, we presented a case of a giant transgeniohyoid cyst extending to the hyoid bone level and demonstrated that the cyst can be totally excised without any complication via an intraoral approach with decompression.

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