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Giant Sublingual Epidermoid Cyst: A Case Report

Gaurav Sapra[1*], Navneet Agarwal[2], Ritu Agarwal[3], Sony Singh[4]
[1]Senior Resident, Department of E.N.T and Head & Neck Surgery, M.D.M Hospital, Dr. S.N. Medical College, Jodhpur, Rajasthan, India
[2]Professor, Department of E.N.T and Head & Neck Surgery, M.D.M Hospital, Dr. S.N. Medical College, Jodhpur, Rajasthan, India
[3]Associate Professor, Department of Anatomy, Government Medical College, Pali, Rajasthan, India
[4]Post Graduate Resident, Department of Microbiology, Dr. S.N. Medical College, Jodhpur, Rajasthan, India

Case Report

***Corresponding author**
Gaurav Sapra

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Abstract: Epidermoid cysts of the floor of the mouth are rare lesions thought to be caused by entrapment of epithelial remnants during embryonic fusion. We present the case of large epidermoid cyst with an oral and a submental component in a 71-year-old male who presented with complaints of a mass in the oral cavity and difficulty in chewing and swallowing solid foods for about 2 years. C.T. Scan shows a 110x55x70mm well-circumscribed cystic mass extending from the sublingual area to the mylohyoid muscle. Extra oral approach was used as the tumor was bulky, whole cyst was removed in toto. We are presenting this case because of the giant size of the cyst and the dilemma in surgical approach to the cyst.

Keywords: Sublingual, Epidermal Cyst, Extra Oral Approach.

INTRODUCTION

Sublingual epidermoid and dermoid cysts are benign lesions encountered throughout the body, with 7% occurring in the head and neck area and 1.6% within the oral cavity [1]. It is defined as "A simple cyst lined with stratified squamous epithelium and lumen is filled with cystic fluid or keratin and no other specialized structure" [2]. The contents of the cyst lining determine the histological categories of the cyst: epidermoid, if epidermis is lining the cyst; dermoid, if skin adenxa exist; or teratoid, if there are tissues derivated from the three germinal layers [3]. Clinically, they usually present as a painless swelling in the floor of the mouth, frequently of a doughy consistency, that may cause difficulty in eating, speaking, and breathing.

An ultrasound scan is commonly used as the first choice to investigate the lesion [4]. The treatment of epidermoid cysts of the floor of the mouth is surgical; the approach can be either intraoral or extraoral, depending on the localization and size of the mass. Cysts are classified into three types by localization: (1) sublingual, (2) submental, and (3) submandibular cysts. Oral approach is usually applied for small sublingual cyst. The extraoral incision is preferred in submental and large sublingual cysts [1]. This report presents a case of large epidermoid cyst of the floor of the mouth in a 51 year-old patient and a review of all steps necessary for diagnosis and treatment.

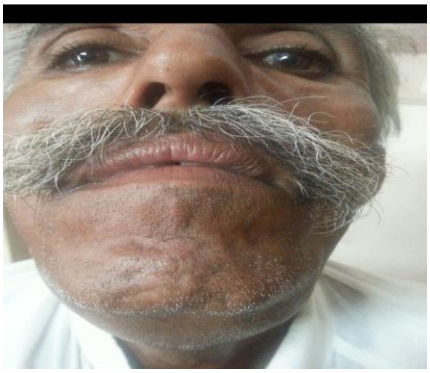

Fig-1: Showing swelling submandibular region extending from one side angle of mandible to other side

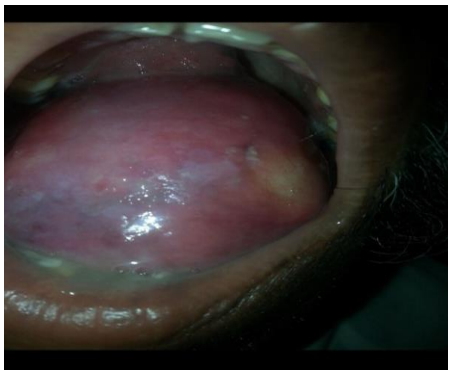

Fig-2: Showing cystic lesion 5x4 cm arising from floor of mouth and pushing the tongue creating a ‘‘second tongue’’ affect.

CASE REPORT

A 51-year-old male patient came to our oto-rhino-laryngology department with the chief complaint of a swelling below the tongue producing difficulty in chewing and swallowing of solid foods for about 2 years. Recently he noticed difficulty in breathing and sought medical attention for the same. Examination revealed the presence of a solitary midline swelling in the neck measuring 6 × 5cm extending from one side angle of mandible to other (Fig 1). It was nontender, fluctuant, soft, and nonmobile, and the overlying skin was normal. There were no inflammatory signs or lymphadenopathy associated with the swelling. Intra-oral examination found a cystic lesion 5x4 cm arising from floor of mouth and pushing the tongue posteriorly and pressing the tongue against the palate, creating a ‘‘second tongue’’ affect (Fig 2). Cross fluctuations between the intraoral and neck swelling were present. The overlying mucosa was normal.

Fig-3: Showing a large cystic space occupying lesion in sublingual region and floor of mouth causing gross compression over tongue (Axial view).

Fig4: Showing a large cystic space occupying lesion in sublingual region and floor of mouth causing gross compression over tongue (Sagittal view).

The C.T Scan showed a large cystic space occupying lesion 110x 55x 70 mm in size in sublingual region and floor of mouth causing gross compression over tongue which was compressed and stretched over the superior aspect of the lesion (Fig-3 & 4). The SOL was extending in FOM inferiorly along the median marring of myelohyoid muscle and extending to submental and submandibular region. Differential diagnosis was plunging ranula, dermoid cyst, cytic hygroma, thyroglossal cyst, ectopic thyroid or lipoma. But looking at the CT finding and the midline position of lesion we felt the lesion was a dermoid. The challenge before the operating team was to decide the approach for surgery and remove the cyst in toto. The size of the lesion being more than 6cm motivated us to go for an Extraoral approach with the possibility of using the combined extra and intra oral approach if required during the surgery. The reluctance of the team to directly use the combined approach was based on possibility of fistula formation post operatively (as the mylohyoid muscles on CT were pushed apart by the lesion) hence the combined approach was kept only to be used if felt need during surgery.

Fig-5: Showing extraoral approach was taken as the tumor was more than 6cm in diameter

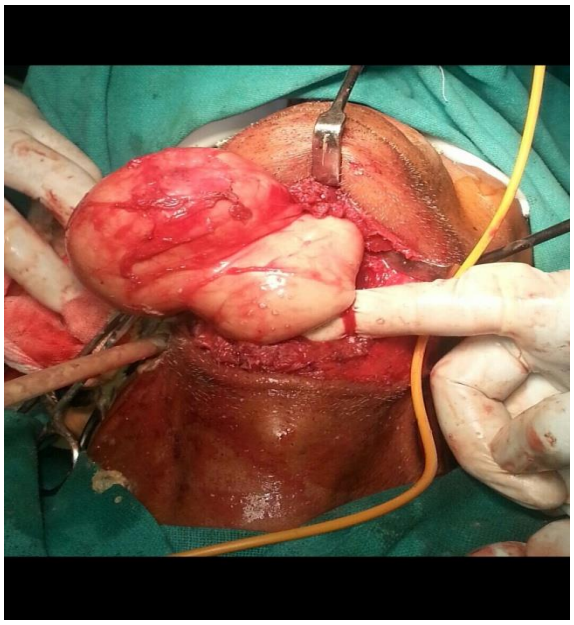

Fig-6: Showing full cyst was removed in toto

The patient was taken for surgery under general anesthesia with nasotracheal intubation. An extraoral approach was taken as the tumor was more than 6cm in diameter, a transverse crease incision was taken, flaps were raised on both the sides, cyst was identified and cyst was seen going intraorally crossing mylohyoid medially, a combination of sharp and blunt dissection was used to free the cyst with traction and counter-traction and full cyst was removed in toto. The wound was closed in layers and a drain was kept in situ for 48 h. Extubation was performed without complications. Clear fluids were started after 48 hrs. No significant edema was observed and healing was uneventful and the patient was discharged on the 7th day after stitch removal.

DISCUSSION

Epidermoid and dermoid cysts are rare, benign lesions found throughout the body, with 7% occurring in the head and neck area, 1.6% of which occurs in the oral cavity. Of all the oral cysts, dermoid and epidermoid cysts account for only 0.01% [5].In 1955, Meyer updated the concept of dermoid cyst to describe three histological variants: the true dermoid cyst, the epidermoid cyst and the teratoid variant. True dermoid cysts are cavities lined with epithelium showing keratinization and with identifiable skin appendages such as pilous follicles, and sudoriparous and sebaceous glands on the cyst wall. Epidermoid cysts are lined with simple squamous epithelium with a fibrous wall and no attached structures. The lining of teratoid cysts varies from simple squamous to a ciliate respiratory epithelium containing derivates of ectoderm, mesoderm and/or endoderm. All three histological types contain a thick, greasy-looking material [6].

Most patients with epidermoid cyst are in the range between 10 and 35 years of age [4]. Growth of the cyst may be increased by hormonal stimulus during puberty, producing a hypersecretion of fat, which would explain the greater incidence in the young adult stage (16-40 years of age) [6].The size and the location of the epidermoid cyst are the cause of the clinical manifestations. Cystic lesions developing above the mylohyoid muscle have the potential to displace the tongue toward the palate and subsequently create difficulty with mastication, speech, and possibly breathing. Cysts developing below the mylohyoid often produce a submental or submandibular swelling [7]. Other forms of mass may confuse diagnosis: thyroglossal tract cyst, cystic lymphangia, ectopic thyroid gland, lipoma, cellulitis, or tumor of the floor of the mouth. Definitive diagnosis is provided by the histological specimen. Imaging can assist diagnosis. Ultrasound reported a pseudo solid an echogenic or finely echogenic cystic mass with fat/liquid line with more echogenic supernatant fluid and/or more echogenic floating mass are suggestive. CT showing a thin-walled unilocular mass, with fatty content. A low-lying liquid line, moving with change in position, is characteristic. MRI finds a fatty level in hyper signal on T1-weighted sequences, with fall in signal on Fat Sat sequences. CT and MRI identify contiguous organs, guiding the choice of surgical approach [8].

Treatment is by enucleation via an intraoral or extraoral approach. An intraoral approach is recommended by most authors for sublingual cysts of small or moderate dimensions (less than 6 cm) above the mylohyoid muscle, whereas an extraoral approach is preferred for larger sublingual cysts (more than 6 cm) [9, 10]. Some authors have also advocated a combined intraoral and external approach for giant dermoid cysts

.In present case, the cyst was large (11.5 cm) and extending mylohyoid muscle, so extraoral approach was used. Recurrence is very rare with complete excision of the lesion, but a 5% rate of malignant transformation has been reported [11].

CONCLUSION

Epidermoid cysts of head and neck origin are quite a rare entity. Appropriate imaging techniques are necessary to look for the extension of the cyst in giant cysts. The surgical approach is made according to the location of the lesion in facial spaces and recurrence after total enucleation is very rare.

REFERANCES

1. Ohta N., Watanabe T., Ito T., Kubota T., Suzuki Y., Ishida A., Kakehata S., & Aoyagi M. (2012). A Case of Sublingual Dermoid Cyst: Extending the Limits of the Oral Approach. Hindawi Publishing Corporation Case Reports in Otolaryngology, Article ID 634949: 4 pages
2. Shafer, W. G., Hine, M. K., & Levy, B. M. (1993). A text book of Oral Pathology. 4th ed. Philadelphia: WB Saunders, p. 67.
3. King, R. C., Smith, B. R., & Burk, J. L. (1994). Dermoid cyst in the floor of the mouth. Oral Surg. Oral Med. Oral Pathol., 78**:** 567–576.
4. Lima, S. M., Chrcanovic, B. R., de Paula, A. M., Freire-Maia, B., & de Souza, L. N. (2003). Dermoid cyst of the floor of the mouth. Scientific W J, 3:156-162.
5. Janarthanam, J., & Mahadevan, S. (2012). Epidermoid cyst of submandibular region. Journal of Oral and Maxillofacial Pathology, 16 (3):435-437.
6. Vargas Fernandez J. L., Lorenzo, R. J., Aneiros F. J., & Sainz, Q. M. (2007). Dermoid cyst of the floor of the mouth. Acta Otorrinolaringol Esp, 58:31-33.
7. Louis, P. J., Hudson, C., & Reddi, S. (2002). Lesion of the floor of the mouth. J oral Maxillofac Surg, 60:804-807.
8. Boko, E., Amaglo, K., & Kpemissi, E. (2014). A bulky dermoid cyst of the floor of the mouth. European Annals of Otorhinolaryngology, Head and Neck diseases, 131:131—134.
9. El-Hakim, I. E., Alyamani, A. (2008). Alternative surgical approaches for excision of dermoid cyst of the floor of mouth. International Journal of Oral and Maxillofacial Surgery, 37(5):497–499.
10. Eken, M., Evren, C., Sanli, A., & Bozkurt, Z. (2007). An alternative surgical approach for sublingual dermoid cysts: a case report. Kulak Burun Boˇgaz Ihtisas Dergisi, 17(3): 176–178.
11. Agaimy, A., Raab, B., Bonkowsky, V., & Wunsch, P. H. (2007). Intestinal-type adenocarcinoma arising in a congenital sublingual teratoid cyst. Virchows Archiv, 450(4):479–481.