REVIEW ARTICLE

Sublingual epidermoid cyst in young female: a case report

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Abstract

Dermoid and epidermoid cysts are developmental pathologies that occasionally occur in the head and neck region. Sublingual space is bounded superiorly by oral cavity mucosa, laterally by the mandible, and inferiorly by geniohyoid and mylohyoid muscles. Dermoid and epidermoid cysts are occasionally found in the sublingual space, despite being frequently found in the paediatric head and neck region. Approximately 7–8% of dermoid cysts appear in the head and neck, but only 1.6% of dermoid cysts involve the floor of the mouth. We hereby present a rare case of epidermoid cyst which was present in the floor of mouth in young female.

INTRODUCTION

Dermoid and epidermoid cysts are developmental pathologies that occasionally occur in the head and neck region. Sublingual space is bounded superiorly by oral cavity mucosa, laterally by the mandible, and inferiorly by geniohyoid and mylohyoid muscles.¹ Dermoid and epidermoid cysts are occasionally found in the sublingual space, despite being frequently found in the paediatric head and neck region.

Approximately 7-8% of dermoid cysts appear in the head and neck, but only 1.6% of dermoid cysts involve the floor of the mouth.^{2,3} Epidermoid cysts are exceedingly rare, accounting for less than 0.01% of all oral cavity cysts.⁴ Most clinicians and researchers believe that dermoid and epidermoid cysts that appear in the midline floor of the mouth are a result of entrapped ectodermal tissue of the first and second branchial arches, which fuse during the third and fourth weeks of intrauterine life. A second theory suggests that midline dermoid and epidermoid cysts may be a variant of the thyroglossal duct cyst with ectodermal elements predominating.⁵ According to the location and size of the lesion, surgical treatment for epidermoid cysts on the floor of the mouth may be intraoral or extraoral. We hereby present a rare case of epidermoid cyst which was present in the floor of mouth in young female

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Case Report

A 09-year-old female patient with moderate build reported to our department with a chief complaint of swelling below her tongue since birth which kept on gradually increasing till present size. The patient also has had difficulty in mastication and deglutition for the last 1 year. There were no significant extraoral findings. Intraoral findings revealed firm painless swelling present in the floor of the mouth which was non-movable and dough-like. The color and texture over the swelling were unchanged (Fig 1). Needle aspiration was found to be negative.

Radiographic investigation computed tomography was done which was suggestive of a well-defined circumscribed mass above the level of mylohyoid muscles. No sign of cortical expansion was evident. (Fig 2,3). From these clinical and radiographic we made a provisional diagnosis of the epidermoid cyst and a treatment plan of excision under general anaesthesia was made. Under all aseptic conditions, an elliptical incision was given on the floor of the mouth intraorally (Fig 4). Complete in-toto excision of the cyst was done which measured about 5 x 3 cm (Fig 5,6). The specimen was further sent for histopathological evaluation.

Histopathologic findings show ortho keratinized stratified squamous epithelium overlying a fibro cellular connective tissue stroma. The underlying connective tissue shows loosely arranged collagen fibers in association with large cystic spaces filled with desquamated keratin and infiltration of inflammatory cells predominantly consisting of lymphocytes (Fig 8). These features are clearly suggestive of Epidermoid Cyst.



Fig 1. Intraoral clinical findings



Fig 2, 3. CT scan sections depicting circumscribed mass



Fig 4. Elliptical Incision





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Fig.7 Excised tissue specimen in 10% buffered formalin



Fig 8. Photomicrograph of the section shows orthokeratinized stratified squamous cystic epithelium without any rete ridges formation (H & E stain at 10x magnification)

DISCUSSION

Epidermoid and dermoid cysts are rare, benign lesions found throughout the body. Roser was the first to designate dermoid cysts in the floor of the mouth as epidermoid tumors.⁶ New and Erich (1937) reported 24 (1.6%) epidermoid cysts occurring at the floor of the mouth out of 1495 cases of dermoid cysts seen at the Mayo Clinic.⁷

In 1955, Meyer updated the concept of the dermoid cyst to describe three histological variants: the true dermoid cyst, the epidermoid cyst, and the teratoid variant.

1. Epidermoid cysts – where the cystic cavity is lined with epithelium without skin appendages.

2. Dermoid cysts – here the epithelial lined cystic cavity encloses skin appendages such as hair, hair follicles, sebaceous, and sweat glands.

3. Teratoid cysts – in this entity, the cystic cavity in addition to skin appendages also encloses mesodermal derivatives such as bone, muscle, gastrointestinal and respiratory tissue.

Epidermoid cysts are lined with simple squamous epithelium with a fibrous wall and no attached structures. 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.⁸ Ambiguity about their exact pathogenesis exists and dysontogenetic, traumatic, and thyroglossal anomaly theories have been postulated.^{9,10,11} Most congenital dermoid and epidermoid cysts perhaps begin due to an embryologic accident during the early stages of development but hardly get perceived until their size causes annoyance.¹² The origin of epidermoid cysts is believed to be from the entrapment of epithelial remnants during midline closure of the bilateral first and second branchial arches.^{13,14} It has also been opined that ectodermal differentiation of multipotential cells, most probably pinched off at the point of anterior neuropore closure may give rise to these cysts.⁹ On the other hand, they may also crop up from the tuberculum impar of His.⁴ Traumatic or iatrogenic inclusion of epithelial cells or the blockage of a sebaceous gland duct has been postulated as the pathogenesis of acquired cyst.^{4,14}

However, some authors have also stated that midline cysts may represent a diverse form of thyroglossal duct cyst. They may be found in any age group but show preponderance between 15-35 years of age with no gender predilection.^{4,9,13} Our reported case is a young female which is very rarely reported in the literature.

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.⁸ Surgical enucleation is the only effective treatment for these kinds of lesions. Several techniques are reported in the literature, which may be divided into intraoral and extraoral techniques depending on which approach is used

In the current case, excision was achieved without major complications by employing intraoral access under general anesthesia.

Under aseptic conditions, elliptical incision was made on the floor of the mouth, followed by blunt dissection. The lesion was found sitting on top of the genioglossus muscle. Complete excision of the cyst was done and further sent for histopathological evaluation which revealed it as an epidermoid cyst. The prognosis is very good, with a very low incidence of relapse

CONCLUSION

Proper clinical evaluation and investigations like CT scans are significant in diagnosing such rare pathologies. The intraoral approach for excision is the best treatment option available in terms of surgical efficiency, aesthetics, and postoperative improved quality of life.

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