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Dysontogenetic cyst – report of an intraoral choriostoma with dermal adnexa.

[1]Dr. Shruti Srinivasan, Senior Lecturer, Department of Oral and Maxillofacial Pathology, The Oxford Dental College, Bengaluru -560068.

[2]Dr. Leeky Mohanty, Head & Professor, Department of Oral and Maxillofacial Pathology, The Oxford Dental College, Bengaluru -560068.

[3]Dr. Seema M, Senior Lecturer, Department of Oral and Maxillofacial Pathology, The Oxford Dental College, Bengaluru -560068.

[4]Dr. Saleha Jamadar, Reader, Department of Oral and Maxillofacial Pathology, The Oxford Dental College, Bengaluru -560068.

[5]Dr. Komali Y, Reader, Department of Oral and Maxillofacial Pathology, The Oxford Dental College, Bengaluru -560068.

Corresponding Author: Dr. Shruti Srinivasan, Senior Lecturer, Department of Oral and Maxillofacial Pathology, The Oxford Dental College, Bengaluru -560068.

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Abstract: Epithelial cells that are entrapped at the sites of fusion of the embryonic processes can proliferate to form cysts. These cysts are called dysontogenetic since they arise due to defective embryonic development. Dermoid cysts are dysontogenetic cysts which can occur at various sites in the body with 7% occurring in the head and neck region and 1.6% in the oral cavity. True dermoid cysts are choriostomas which clinically mimic other pathologies depending on the presenting site but can be distinguished by their unique histopathological features which can be attributed to the ectoderm origin of these entrapped cells. We here report a case of true dermoid cyst of the floor of the mouth at the midline clinically diagnosed as ranula.

Keywords: Dermoid cyst, Dysontogenetic cyst, Choriostoma, Ranula.

Introduction

Dermoid cysts are rare developmental malformations arising from the epithelial cells that get entrapped due to defective embryonic development (Dysontogenetic) and also as a result of traumatic implantation into deeper tissues; hence can be congenital or acquired respectively.[1] These entrapped cells are pluripotent and diffentiate to form dermal adnexal components like

sebaceous gland, hair follicle and sweat glands in their wall.[2] These cysts usually present as midline lesions especially in ovary, testis and sacral region but may occur elsewhere as well.[3,4]

In the head and neck, lateral eyebrow is the most common site followed by floor of the mouth accounting for about 26% of the head and neck dermoids.[4] The other sites in the oral cavity include lips, tongue, buccal mucosa, palate, uvula, parotid gland and jaw bones.[5] Dermoid Cysts of Floor Of Mouth (DCFOM) are usually congenital and appear as midline swellings.[6] These may involve sublingual (52%), submental (26%) or submandibular (16%) region.[5]

Dermoid cysts present at any age but most commonly seen in 2nd to 3rd decade of life with no gender predilection.[7] These usually develop insidiously with patient not becoming aware until they are large enough to cause difficulties in eating and swallowing.[7] They appear as firm swellings which are doughy in consistency.[7] These swellings may grow to variable size ranging from few mms to cms.[2] Those at the floor of the mouth can be misdiagnosed clinically as various other pathologies but can be diagnosed by their histopathological features which consist of an epithelial lining and dermal adnexal structures like sebaceous gland, hair follicles and sweat glands in their cyst wall and abundant keratin in their lumen.

We here present a rare case of a true DCFOM at midline which was clinically diagnosed as ranula.

Case report

A 21yr old female patient complained of swelling below the tongue since 5 years. Patient experienced difficulty in mastication and speech and hence presented to the hospital (Fig -1). An ultrasound was done prior to her visit to the dental hospital which showed a lesion with distinct borders measuring 2.1 x 2.3 cms. Further details of the scan were not available with the patient.

On examination, a solitary median swelling occupying the entire floor of the mouth causing elevation of the tongue was noted. The mucosa over the swelling appeared stretched and was of same colour as the adjacent mucosa. On palpation the inspectory findings were confirmed; the swelling was painless, soft and fluctuant in consistency. Regional lymph nodes were not palpable. A clinical diagnosis of ranula was made based on the findings. Complete enucleation of the lesion was done and the specimen was sent for histopathologic evaluation.

On gross examination, the lesion appeared roughly oval measuring 3.9 X 3.3 X 2.2 cms and appeared whitish cream in colour. (Fig – 2) The cut surface showed cheesy thick yellowish white material which fills the entire lesion. (Fig – 3)

Histopathology showed cystic lining comprising of stratified orthokeratinised epithelium with the thin fibrovascular wall showing dermal appendages like sebaceous gland and hair follicles. Abundant keratin was seen in the lumen. A diagnosis of dermoid cyst was made based on the clinical and histopathological findings. (Fig – 4,5)

Discussion

Dermoid cyst should be the clinical term for all the developmental cysts in the floor of the mouth as suggested by Meyer.[6] Hence, DCFOM comprise a group of benign, developmental malformations which form a spectrum of histopathologic features. These include true dermoid cysts, epidermoid cyst and teratomata / teratoid cysts.[7] Dermoid cysts are thought to be cystic teratomas which do not show derivatives from all the three germ layers.[7] These developmental cysts at the floor of the

mouth can be distinguished from one another only by their histopathology.

Dermoid cysts are usually congenital but may also be acquired. Midline congenital dermoid cysts are formed primarily from failure of separation of ectoderm from neural tube during 3 – 5weeks of intrauterine life. These can also form as a result of sequestration of ectoderm at other sites of fusion of embryonic processes.[8] These are most commonly seen in ovary and testis. The lateral dermoid cysts may arise from the first pharyngeal pouch or the branchial cleft.[8] Acquired cysts form due to implantation of ectoderm into deeper tissues due to trauma or maybe iatrogenic or from the occlusion of sebaceous gland duct.[8]

Various classification systems for DCFOM have been proposed, Meyer (1955) – Based on Biologic types of cysts, Seward (1965) – Based on location in relation to muscles and Spouge (1973) – Based on histopathological features, but none of them gained universal acceptance.[9]

Dermoid cyst of the floor of the mouth can occur in sublingual, submental region.[5] Sublingual dermoid cysts are the most common site and are seen on the floor of the mouth as midline swellings which elevate the tongue causing dysphagia, dysarthria, dysphonia. If occurring in submental region, appear as midline swelling which may give a double chin appearance.[5]

Clinically these must be differentiated from other swellings occurring in the floor of the mouth such as ranula, thyroglossal duct cyst, swelling due to blockage of Wharton's duct, infections of salivary glands, benign and malignant tumours of the floor of the mouth and adjacent salivary glands.[10]

Imaging techniques for swelling in the floor of the mouth include MRI, CT scan and Ultrasound. All these techniques reveal a unilocular, hypodense lesion with distinct borders. MRI of a dermoid cyst of the floor of the mouth may reveal intracystic floating corpuscles which give a "Sack of Marbles" appearance which is pathognomic of these lesions.[5]

Microscopically these cysts are a squamous epithelial lining with dermal appendages like sebaceous gland, hair follicle and sweat gland in their wall. The lumen is filled with keratinous debris.

These rarely recur if excised completely.[7] Malignant transformation in dermoid cyst is rare (less than 2%) with the most common malignancy being squamous cell carcinoma followed by adenocarcinoma.[11]

Conclusion

Dermoid cysts at the midline of floor of the mouth are rare and hence are least thought of in the differential diagnosis of swellings in this region. They clinically resemble other lesions but microscopically show unique features that are not native to the local tissue and thus help in easy diagnosis.

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Legend Figures

Fig 1: Clinical Picture: Median solitary swelling at the floor of the mouth causing tongue to be elevated.

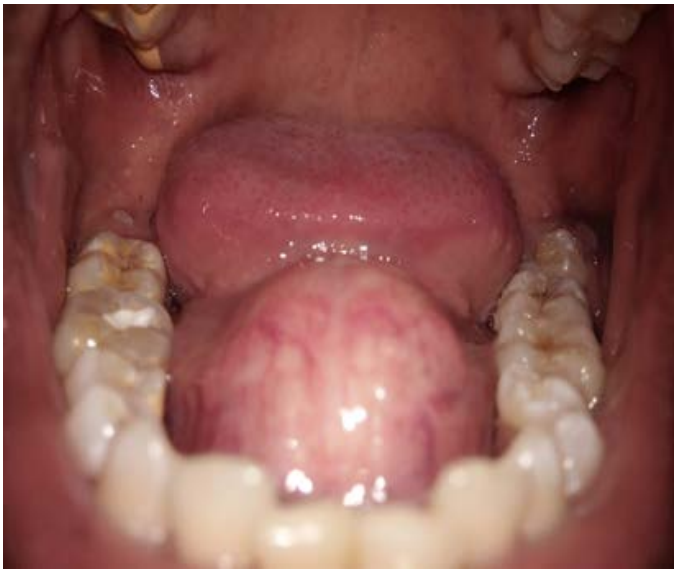

Fig 2: Gross Picture: The excised specimen appears roughly oval and creamish white in colour. Measures about 3.9 X 3.3 X 2.2 cms.

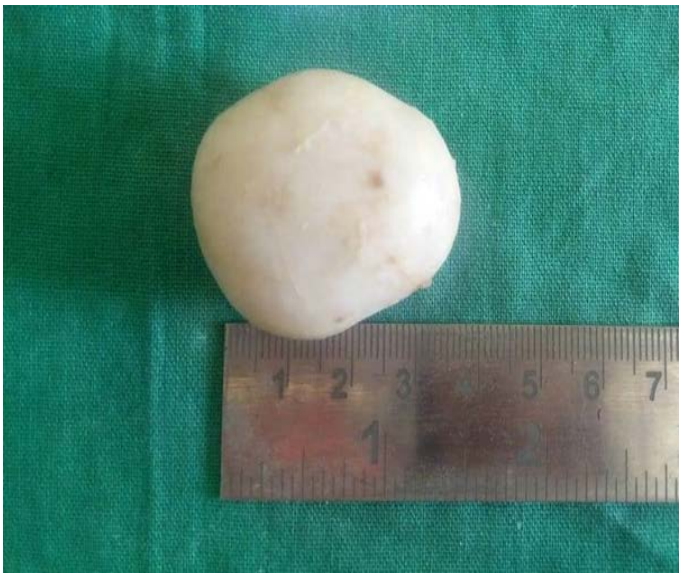

Fig 3: Gross Picture 1: The cut surface shows thick creamy material filling the entire specimen.

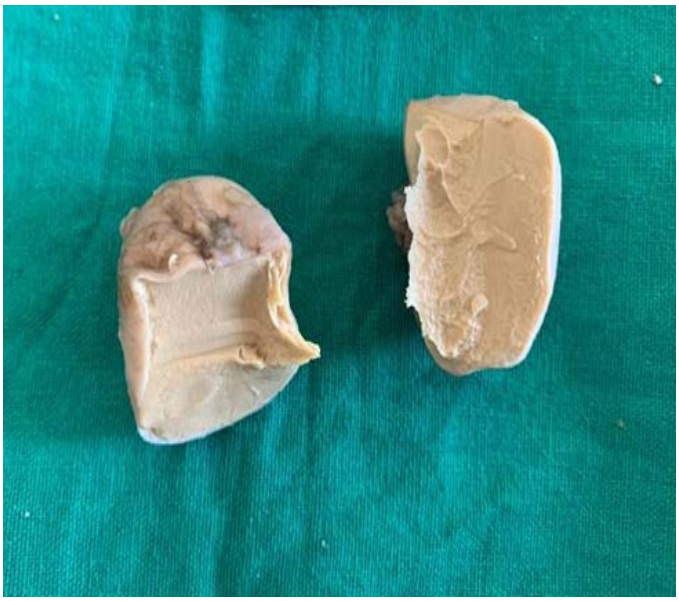

Fig 4: Histopathological Picture: H & E-Stained section showing Orthokeratinised stratified squamous epithelium with fibrovascular wall showing hair follicle. Abundant keratin can be seen in the lumen.

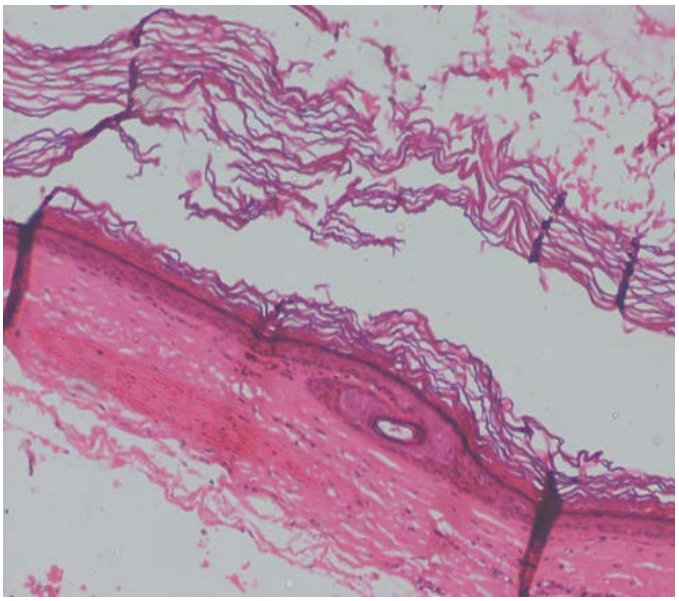

Fig 5: Histopathological Picture: H & E-Stained section showing cystic wall with sebaceous gland component.