Unusual Recurrent Dermoid Cyst: A Case Report

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Dermoid cysts are unusual developmental lesions occurring most frequently on the face, scalp, neck, and trunk, and represent <0.01% of all cysts of the oral cavity. Histologically, depending on the presence of adnexal structures and derivatives of germ layers, these cysts are classified as dermoid, epidermoid, and teratoid cysts. Anatomically, they are divided into median genioglossal, median geniohyoid, and lateral cysts. Clinically, a distinction between supra and inferior type as well as between central and lateral type is proposed in relation to the mylohyoid muscle and the midline, respectively. Enucleation via intraoral and/or extraoral approach is the method of treatment. This cyst elicits a good prognosis with low malignancy and recurrence rate.

Keywords: Dermoid, Hamartomatous, Multipotent

INTRODUCTION

Dermoid cysts are rare developmental benign lesions that arise as a result of ectodermal differentiation of multipotent cells. This hamartomatous lesion basically arises from entrapped midline ectodermal tissue lined by epidermis with skin appendages present in the fibrous wall. Epidermoid and dermoid cysts may occur anywhere in the body, but most predominantly in the ovary and scrotal regions. The floor of the mouth is one of the most commonly affected areas, however, these cysts can also be found in the tongue, lips, buccal mucosa, and jaw bones.

Dermoid cysts commonly affect people between the ages of 15 and 35 years. Lesions are usually asymptomatic, slow-growing, and are a well-encapsulated mass with a doughy like feel on palpation. These cysts have a good prognosis.

Dermoid cyst is an uncommon lesion of developmental origin, lined by squamous epithelium containing desquamated cells. Jordan in 1778 gave the term sublingual dermoid cyst. In 1838, Lawrence identified a palatal dermoid cyst for the first time in literature. In 1955, Meyer gave three histological variants: Dermoid, epidermoid, and teratoid cyst.

Only 7% of cases are reported in the head and neck area with the common site being the lateral eyebrow followed by the floor of the mouth. In the orofacial region, the median site, paramedian site, suprhyoid region, and lateral site are most common.

Histologically, this distinction of the cysts in the floor of the mouth was presented by Meyer in 1955. The cyst is described as epidermoid when the lining presents only epithelium, dermoid when skin adnexa are found, and teratoid when other tissues, such as muscle, cartilage, or bone, are present within the cyst.

The treatment of dermoid 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.

In this case report, we describe a case of recurrent dermoid cyst in a patient occurring in the left mandibular angle region.

CASE REPORT

A 45-year-old male patient reported to the dental hospital outpatient department (OPD) with a chief complaint of pain in left lower back region of face since 4 days. Patient was apparently asymptomatic 4 days back when he had a fall while walking.

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Pain was spontaneous, localized, aggravated on jaw movement, and was relieved on taking medication. No history of hypertension or diabetes was given by the patient. Patient was treated for mandibular angle trauma at a private dental clinic.

Patient was moderately built, conscious, and cooperative. Extraoral examination revealed a well-defined, painless, solitary swelling measuring 3 cm × 4 cm in size at the site of fracture (mandibular angle area). The extension of the swelling was anteroposteriorly, a line drawn from inner canthus of the eye to 1 cm ahead of the tragus of the ear. Superoinferiorly it was extending from the left corner of the mouth to the tragus of ear and line drawn along the lower border of the mandible. The swelling was non-pulsatile and oval in shape having normal overlying skin. On palpation, the surface of the swelling was smooth, non-tender, compressible, soft in consistency, and mobile (not attached to the underlying tissues). No intraoral swelling was detected.

On the basis of clinical presentation, a provisional diagnosis of the dermoid cyst was given with differential diagnosis of lipoma and sebaceous cyst. Cystic enucleation was done under local anesthesia. Patient was treated for mandibular angle fracture. Open reduction with internal fixation was done under general anesthesia. The resected specimen was sent for histopathological examination. Gross features revealed a single piece of soft tissue measuring 1.0 cm × 0.5 cm, firm in consistency, black in color, and smooth surface with attached hair follicle.

Histopathological examination revealed a cystic lining of stratified squamous epithelium and filled with desquamated keratin in a laminar pattern. Underlying connective tissue stroma was dense, fibrous, and showed few inflammatory cell infiltrate, hair follicles, extravasated red blood cells (RBCs), and hemosiderin pigment (Figure 1). A daughter cyst was also seen with keratin-filled cystic lumen (Figure 2). On the basis of these features, a final diagnosis of the dermoid cyst was signed out.

The same patient came after 2 months to the hospital OPD with a chief complaint of a swelling in the left lower back region of the face since 1-month.

On extraoral examination, a well-defined, painless, solitary swelling measuring 2 cm × 3 cm in size was found at the original site (Figure 3). The swelling was non-pulsatile and oval in shape having normal overlying skin. On palpation, the surface of the swelling was smooth, non-tender, compressible, soft in consistency, and mobile (not attached to the underlying tissues) in nature. No intraoral swelling was detected. On the basis of clinical presentation, a provisional diagnosis of the dermoid cyst was given.

Cystic enucleation was done under local anesthesia. Gross features revealed a single piece of soft tissue, yellowish brown in color, soft in consistency, and irregular in shape measuring 1.2 cm × 1.5 cm.
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Histopathologic examination revealed cystic lining of stratified squamous epithelium filled with desquamated keratin in the laminar pattern (Figure 4). Underlying connective tissue stroma was dense, with sparse inflammatory cell infiltrate, sebaceous glands, extravasated RBCs, and hemosiderin pigment (Figure 5).

On the basis of this feature, a final diagnosis of the recurrent dermoid cyst was given.

**DISCUSSION**

Dermoid cysts are unusual clinicopathological lesions which result from defective embryonic development. Based on their origin, they are classified as acquired and congenital. Three theories have been proposed regarding the origin of the cyst.

1st theory states that the origin is from the embryonic cells of 1st and 2nd branchial arches entrapped in the mesenchyme during 3/4th few weeks of embryonic life. 2nd theory explains various acquired cysts due to the implantation of the epithelial cells subsequent to any injury or occlusion of sebaceous gland duct. According to the 3rd theory, they are considered as a variation of the thyroglossal cyst.

Dermoid cysts may be found in any age group. Highest incidence of the cyst is between 15 and 35 years of age. Women are affected more than men (5 of the 7 patients were women, i.e., 70%).

Clinically, dermoid cysts present as a slow growing mass so they are diagnosed only after having reached a considerable size (2-5 cm). They may be fluctuant and cyst-like based on the consistency of the luminal contents which may be cheesy, sebaceous or liquefied substance. The cyst is non-reducible with bi-digital palpation. The cysts show negative transillumination and fluctuation positive.

Diagnostic imaging of the lesion includes computed tomography (CT) scan, magnetic resonance imaging, and ultrasonography. The use of radiograph or orthopantomogram may not be very useful unless a radiopaque medium is injected into the lesion. On CT scan, the dermoid cyst appears as moderately thin walled, unilocular mass filled with a homogenous fluid with numerous hypoattenuating fat nodules, giving the pathognomonic “Sac of marbles” appearance. The cystic contents are often keratinous, caseous, sebaceous or purulent with hair follicles, fat globules, and cartilage.

Histologically, both dermoid and epidermoid cysts are lined by keratinized squamous epithelium resembling epidermis. Epidermoid cysts do not contain any adnexal structures. Teratoid cysts contain derivatives of mesoderm, ectoderm, and endoderm. The dermoid cyst is characterized by the presence in its wall of one or more dermal appendages such as hair follicles, sweat glands, or sebaceous glands. The lumen is usually filled with keratin. Occasionally, the lining epithelium may proliferate as papillary fronds extending externally or inward toward the cystic lumen. This proliferation may have some superficial resemblance to epidermal carcinomatous proliferation, and the growth may be misdiagnosed as a malignancy.

Surgical enucleation is the treatment of choice for all variants of dermoid cysts with a good prognosis. The relapses can be related to the presence of further dysembryoplastic cell groups or to an incomplete excision of the cyst. It has a good prognosis and shows rare malignant transformation. 5% rate of malignant transformation of oral dermoid cysts into the teratoid type has been reported in literature. Very
rarely, the dermoid cyst invites complications such as suppuration, fistulization, or foreign body reactions due to the break of the wall.\(^4\)

**CONCLUSION**

In this case, the cyst started as a gradually increasing swelling and showed a recurrence in the same region 2 months after surgical excision. Histopathologic examination confirmed the features of dermoid cyst.

Thus, this case highlights the importance of prompt and effective surgical treatment to prevent recurrence of this cyst.

**REFERENCES**