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Research Article

IDIOPATHIC SIALACTESIA LEADING TO MUCOUS RETENTION CYST-A CASE REPORT

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ABSTRACT

'Mucocele' literally means a cystic cavity filled with mucin. It is the most common non-neoplastic salivary gland pathology particularly in the minor salivary glands. There are of two main types of mucocele; retention type and extravasation type. occurs owing to a glandular duct obstruction and resultant accumulation of mucus in duct cavity. The lesion can fluctuate in size, depending on its fluid-filled state. Thus, mucoceles typically present as single, recurrent, painless, well-circumscribed and often bluish nodules.

A 38-year-old male patient reported to the Dental out Patient Department of Government Dental College& Hospital, Raipur with the chief complaint of a swelling in his left posterior cheek region since 10 years. On palpation, the lesion was compressible& fluctuant with watery discharge into oral cavity on compression.

USG was suggestive of Stenson's duct Ectasia(Sacular dilatation).

Gross specimen was an intact saclike mass of 2x3x4 cm which when cut released watery fluid.
HP examination specimen revealed a cystic cavity lined by ductal stratified squamous 3-4 cell thick epithelium and supported by fibrovascular connective tissue capsule.

A final diagnosis was formulated as Mucous Retention cyst of the Buccal space with idiopathic Stenson's duct sialectasia was given.

INTRODUCTION

'Mucocele' literally means a cystic cavity filled with mucin. It is the most common non-neoplastic salivary gland pathology particularly in the minor salivary glands, which usually presents as soft-fluctuant nodular swelling and is generally asymptomatic. There are of two main types of mucocele; retention type and extravasation type.

A mucous retention cyst is a benign pathologic lesion .It occurs owing to a glandular duct obstruction and resultant accumulation of mucus in duct cavity. The lesion can fluctuate in size, depending on its fluid-filled state. A decrease in lesion size is frequently associated with a history of drainage of a thick viscous fluid. The lesion is nonpainful, soft, doughy, and fluctuant to palpation. Clinically, the overlying mucosa may have the same coloration as the lower lip or have a bluish hue. Lesions of longer duration may appear firmer and fibrotic and be difficult to distinguish from a fibroma. A mucocele most likely results secondary to a traumatic event that, in most situations, goes unrecognized. The lower lip is the most common location. However, other sites, including the upper lip and the buccal mucosa, can also be affected. Thus, mucoceles typically present as single, recurrent, painless, well-circumscribed and often bluish nodules.(1,2,3)

A case of large mucus retention cyst of buccal space as a consequence of Idiopathic Sialectasia is described and the pathogenesis and surgical procedure are discussed.

Case Report

A 38-year-old male patient reported to the Dental Out Patient Department of Government Dental College& Hospital, Raipur with the chief complaint of a swelling in his left posterior cheek region since 10 years. He had a H/O gradual increase in the size of the lesion & pain in association with the lesion region.

Patient had an extra-oral swelling extending antero-posteriorly from corner of mouth to angle of mandible region of the colour of skin. Intra-oral examination showed normal left buccal mucosa. A physical examination revealed a large, 3 cm, intraoral palpable mass that was grossly visible on the external left cheek and palpable submucosally intra-orally (Fig 1)

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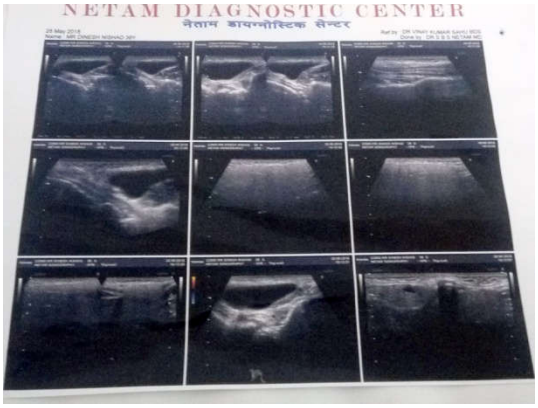
On palpation, the lesion was compressible& fluctuant . On compression of lesion, it was firm but fluctuant & showed watery discharge into oral cavity subsequent to which the pain would be relieved. The cervical lymph nodes were not palpable. He had pain during chewing and speaking.

The patient had no significant family history of similar lesions and had no previous relevant medical history. However, he did have a habit of Gutkha chewing for about 2 years.

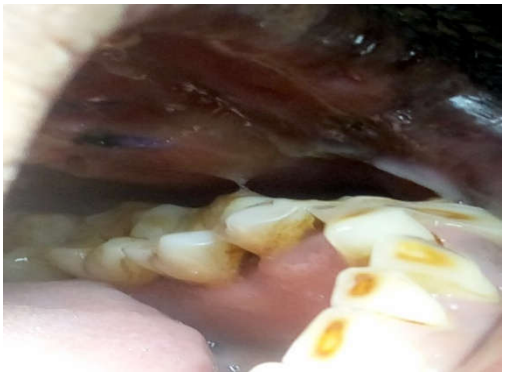
He had noticed that the swelling progressively increased and there was a significant change in the size of the lesion at the time of meals. He did not commit to any incident of trauma or cheek biting in that area prior to lesion development. Based on clinical examination, a provisional diagnosis of Mucocele, with a differential diagnosis of Lipoma was made.

Investigations

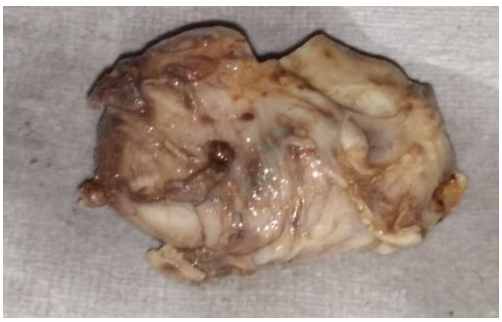
1. Fine needle aspiration cytology (FNAC) was done, and 1 ml of thick, viscous, water colored mucus secretion was collected and sent for chemical analysis which showed increase in amylase and protein content.
2. Ultrasonography revealed a cystic lesion in relation to Stenson's duct. It revealed one clear anechoic(echoless) cystic lesion measuring 34.8 mm×12.2 mm×27.7 mm. It seemed to be communicating with the Stenson's duct . USG was suggestive of Stenson's duct Ectasia(Sacular dilatation).However, Parotid gland & Masseter muscle appeared normal. (Fig. 2)



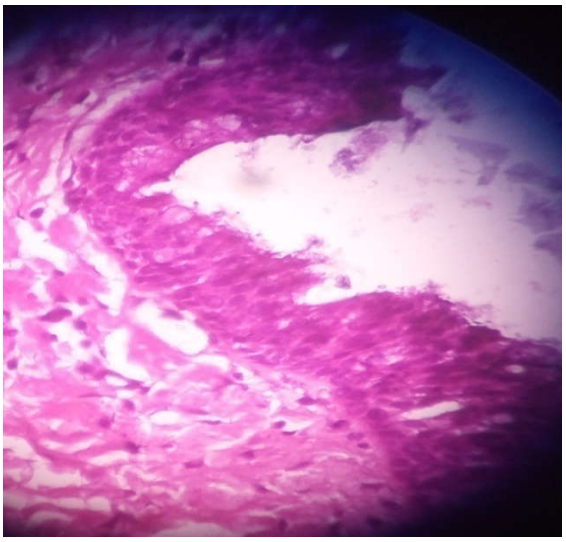
After obtaining the patient's consent an excisional biopsy was done with an Intraoral approach and the specimen was sent for histopathological examination. Complete excision of 2x3x4 cm mass was performed. Submucosal tissue flaps were dissected superiorly and inferiorly to expose the collection in the left buccal mucosa spreading antero - posteriorly by IO surgical procedure. The parotid gland was pushed posteriorly by the cyst but there was no connection between them. Excision of the dilated portion of the duct with stent placement and suturing of the ductal orifice to the buccal mucosa was done with excellent postoperative result.
(Fig 3)

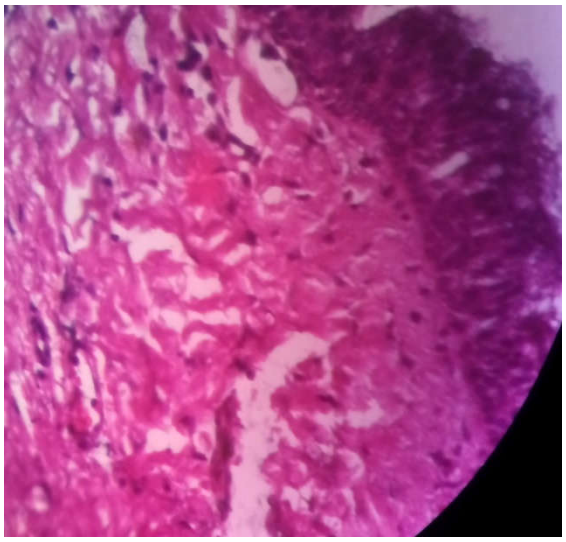


At the time of grossing, two small triangular halves, were taken from the intact saclike specimen & a mucinous , watery, gel-like fluid was seen on the cut surface.



The histopathological examination of the specimen revealed a cystic cavity lined by 3-4 cell thick ductal epithelium and supported by fibrovascular connective tissue capsule (Fig. 3). The lining epithelium was comprised of stratified squamous epithelium (Fig. 4). Some amount of patchy inflammatory cell infiltration was seen.





A final diagnosis was formulated as Mucous Retention cyst of the Buccal space with idiopathic Stenson's duct sialectasia from the clinical features and investigations (chemical analysis, USG, excisional biopsy).

Differential Diagnosis

The differential diagnosis is broad and can include sialocele, underlying sialolithiasis, stricture or mucus plugging. The diffuse form of sialectasis usually presents with a tube-like swelling in the cheek following the route of the Stensen's duct and may be diagnosed using sialography, ultrasonography and CT which will reveal a dilated Stensen's duct without any obvious cause of obstruction. The focal form of sialectasis may present with a soft ballotable facial mass with minimal proximal dilation of the affected duct.(4)

DISCUSSION

According to European and American literature, salivary duct cysts constitute 10% of all cysts of the salivary glands. They may be of congenital or acquired origin. Imaging plays an important role in investigation of salivary duct cysts in delineating the borders, extent of involvement, and the content. Ultrasonography reveals a well-defined lesion with imperceptible walls, anechoic center, and posterior acoustic enhancement. (8)

Focal sialectasis, which is defined as a dilation of a salivary duct, is a rare entity affecting the salivary glands. The disease process is thought to occur as a result of chronic obstruction with dilation. Repeated dilation can lead to a weakening of the ductal wall, which can be either focal or diffuse and results in permanent dilation and enlargement of the ductal calibre.

Idiopathic Dilatation of the Salivary Ducts: This problem is attributable to a stricture or stenosis located at the level of the sphincter of the main salivary duct that causes a dehiscence of the wall of the duct. The dilatation resembles a hernia of the main salivary duct, occurring during meals. As the disease progresses, pain can also occur, giving rise to a salivary colic, and bilateralization generally occurs. At this stage, the duct can be seen and palpated under the skin of the cheek. It occurs most often in Stensen's duct of the parotid gland. (5) There is a condition called Congenital dilatation of Stensen's duct (CDSD) which is a rare heteroplasia of the parotid gland, which may have a hereditary background. It presents as a painless and progressive swelling in the cheek along the Stensen's duct without an obvious cause. The static secretions undergo secondary infection causing pain, fever, and intraoral purulent flow from Stensen's duct. They have a horizontal distention with a unique & peculiar facial swelling somewhat tubular in configuration along the course of Stenson's duct. They usually have a longstanding presence with periods during which they become more prominent. (10)

Combining the literature with their experience, Wang et al (2011) summarized the diagnostic features of CDSD as follows

1. the primary symptom of painless swelling in the cheek that is not related to eating, without any evident etiology;
2. may be unilateral or bilateral, and may occur in any age group;
3. clinically, the presence of swelling along the Stensen's duct (in patients without a history of inflammation, aggressive massage of the swelling can produce abundant intraoral salivary flow); and
4. parotid sialography demonstrates dilated Stensen's duct with a smooth margin but no evidence of obstruction. Our patient seems to fit the above mentioned diagnostic criteria for CDSD.(6,7)

In our case, the dilatation of Stenson's duct had led to stagnation of salivary flow leading to Retention mucocele which is a true,soft tissue cyst of the salivary glands due to decrease or absence of glandular secretion produced by blockage of salivary gland ducts.

Histological investigation paves is essential to final diagnosis and should be carefully evaluated. These lesions are mostly unilocular, lined by ductal epithelium, which may be cuboidal or columnar, and completely or partly lined by squamous epithelium. Sparse to moderate lymphocytic infiltrations may be observed in the cyst wall. Occasionally, oncocytic metaplasia (often seen in cases following ductal obstruction) is seen. The lesions composed of oncocytic cells range from oncocytic metaplasia, and hyperplasia to benign and malignant neoplasms, including oncocytomas and oncocytic carcinomas. (8)

Treatment of sialectasis—such as repeated aspiration, compression, dilation of the papilla and stent placement—may not result in the successful resolution of this condition. When conservative treatment fails—as in the case described here—a more aggressive surgical management may be indicated for distal sialectasis. Though rare this entity needs to be diagnosed timely to prevent recurrent parotitis and treated accordingly. The case presented here represents the first report of such a big retention cyst in buccal space alongwith Stenson's duct Sialactesia. In 1988 Rodgers and Myers pointed that there is an ever-present danger of injury to the facial nerve and Stensen's duct as a result of intraoral surgery of the masses which are localized in buccal space. However, some of the dental surgeons still prefer intraoral extraction of masses in the buccal space. Superficial and minor mucoceles can be removed by intraoral surgery, but it is dangerous to remove bigger mucoceles or masses which placed in lateral or medial part of buccal space by this approach. Masseter, zygomaticus and orbicularis oris muscles should be accepted as anatomical land

marks to choose the approach for surgery. Rhytidectomy approach and extended parotid-submandibular approach are thus,recommended for masses in buccal space.(3,9)

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