Case Report

Salivary Cyst of Floor of Mouth: A Case Report.

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Abstract: The term "ranula" describes a diffuse swelling on the floor of the mouth caused either due to a mucous extravasation or, less commonly, a mucous retention phenomenon. They are generally derived from the major sublingual or submandibular salivary glands. Mucoceles are an extremely common lesion of the minor salivary glands, however a rare occurrence in the major salivary glands is noted. This case report presents an unusual case of a sublingual gland mucocele presenting as a polypoid mass clinically and histopathologically seen as a mucocele. A clinical diagnosis of ranula was made. The term plunging or cervical ranula refers to mucoceles that extend below the mylohyoid muscle, beyond the sublingual space and invariably are associated with the sublingual salivary gland.

Key words: Gland; Mucocele; Ranula; Salivary; Sublingual.

INTRODUCTION

Mucocele are defined as mucus-filled cavities, which can appear in the oral cavity, appendix, gallbladder, paranasal sinuses, and lacrimal sac.[1] Oral mucocele can be of two types: Extravasation and retention. Extravasation mucocele results from a broken salivary gland duct and consequent spillage into the soft tissue around this gland. Retention mucocele appears due to decrease or absence of glandular secretion produced by blockage of salivary gland ducts. Histologically the extravasation type has no epithelial lining and is formed by a mucus pool surrounded by granulation tissue and the retention cyst has an epithelial lining.[2]

Ranulas are mucoceles that occur in the floor of the mouth usually involve the major salivary glands. The term ranula is used because this lesion often resembles the swollen abdomen (belly) of a frog (rana).[3]Ranula originates in the body of the sublingual gland, in the ducts of Rivini of the sublingual gland, and, infrequently from the minor salivary glands at this location. They can be categorized into 2 types: oral ranulas and cervical/ plunging ranulas. Oral ranulas are secondary to mucus extravasation that pools superior to the mylohyoid muscle, whereas cervical ranulas are associated with mucus extravasation along the fascial planes of the neck. Rarely, it may arise within the submandibular gland and presents as a plunging ranula.[4] Here we are reporting one such case of mucocele in the form of oral ranula in a male patient aged 26years.

CASE REPORT

A 26 years old male patient reported to a private clinic with a sizable swelling on the floor of mouth. The lesion was slow growing and there was not associated with any pain. The only symptom presented was a local discomfort and difficulty in mastication. On extraoral examination mild swelling was noted in the left submandibular space area. No other sign of inflammation was present. Intraorally, a solitary, well-defined, dome-shaped, reddish mass was seen of around 2cms in diameter, arising from the floor of the mouth (Fig 1). Clinical diagnosis of a ranula was made and excisional biopsy of the lesion was performed and the tissue was submitted for histopathological examination.

Figure 1: Mucocele seen on the left side of floor of mouth.

On light microscopic examination, the H&E stained sections of the cystic lesion showed atrophic to hyperplastic parakeratinized stratified squamous surface epithelium with underlying connective tissue stroma. Cystic space containing mucin, lined by granulation tissue wall was also evident. Few mucinogens were also evident within the

cystic space. The fibrocellular stroma showed inflammation along with numerous dilated and proliferated blood vessels surrounding the cystic cavity (Fig 2). A confirmatory diagnosis of Mucocele was given.

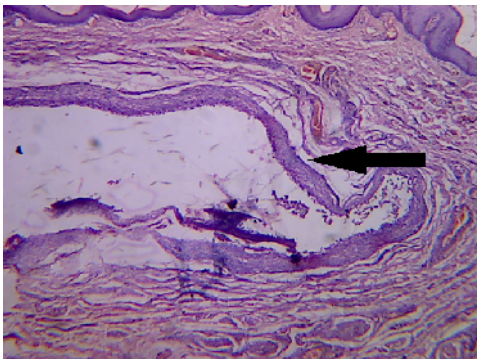

Figure 2: Cystic space lined by granulation tissue wall (thick arrow).

DISCUSSION

Mucoceles (muco - mucus and coele - cavity), are cavities filled with mucus, and are commonly occurring as benign soft tissue masses with a prevalence of 2.4 cases per 1000 people.[2] Clinically may be characterized by single or multiple, painless, soft, smooth, spherical, translucent, fluctuant nodule, which is often asymptomatic.[5]

Mucoceles occurs in young individuals, and affects patients of all ages. The highest incidence of occurrence is second decade of life. Teenagers and children are most commonly affected by mucoceles. Superficial mucoceles tend to occur in individuals older than 30 years and ranulas occur in children and young adults, with a peak frequency in the second decade. Mucus retention cysts, tends to occur in older individuals with the peak prevalence occurs in persons aged 50–60 years.[2] Patient reported here was 26 yrs of age.

Oral mucoceles are said to arise equally in both the sexes, with Male:Female ratio of 1.07:1.[5] Ranula are most common in the second decade of life and predominantly seen in females.[2] They can occur in varying locations on the oral mucosal surfaces overlying accessory minor salivary glands although lower lip is the most frequent site of occurrence which is followed by tongue, floor of mouth (ranula), and the buccal mucosa.[2] The oral mucoceles located on the floor of mouth are termed as '*ranula*', the name is derived from the typical swelling that resembles the air sacs of the frog - 'rana tigrina', which usually arises in the body of the sublingual gland. Occasionally it may arise in the ducts of Rivini or in the Wharton's duct.[6] Ranulas are considered a variant of mucoceles which manifests as a cup-shaped fluctuant bluish swelling on the floor of mouth and tends to be larger than mucoceles located in other regions of the mouth, reaching some centimeters in diameter.[7] Our case was a typical of oral ranula, arising from the floor of mouth but with increased vasularity.

The etiology of oral mucoceles is obscure. Trauma and obstruction of salivary gland ducts are considered crucial factors.[5] The clinical appearance of a mucus cyst is a distinct, fluctuant, painless swelling of the mucosa. About 75% cases shows lesion smaller than 1 cm in diameter; however, the size can vary from few millimeters to several centimeters.[2]

The color ranges from deep blue to the normal color of oral mucosa (pink). Superficial lesions take on a bluish to translucent hue, whereas deep lesions have normal mucosal coloration and bleeding into the swelling may impart a bright red and vascular appearance.[2] The deep blue color may be a result of tissue cyanosis or vascular congestion that is associated with the stretched overlying tissue and the translucency of the accumulated fluid beneath. The variation depends on the size of the lesion, proximity to the mucosal surface and the elasticity of the overlying tissue.[8,9] The swelling was erythematous in our case which could be because of trauma which was giving it a vascular appearance.

The various differential diagnoses considered were Blandin and Nuhn mucocele, oral hemangioma, oral lymphangioma, lipoma, venous varix, irritational fibroma, benign or malignant salivary gland neoplasms, and soft tissue abscess.[2,5] The lesion can be clinically diagnosed as vascular lesion, pyogenic granuloma, polyps or squamous papillomata depending on the degree of vascularity, scarring & acinar atrophy.[7]

The development of mucoceles and ranulas depends on the disruption of salivary flow from the secretory apparatus of the salivary glands. They may be associated with mucus extravasation into the surrounding soft tissues d\t traumatic ductal insult, which could be a crush-type injury and severance of the excretory duct of the minor salivary gland. The disruption of the excretory duct results in extravasation of mucus from the gland into the surrounding soft tissue. Another possible mechanism could be the rupture of acinar structure caused by hypertension from the ductal obstruction. Trauma that results in damage to the glandular parenchymal cells in the salivary gland lobules is another potential mechanism.[4]

Partial or total excretory duct obstruction is involved in the pathogenesis of ranulas.this may be due to a sialolith, congenital malformation, stenosis, periductal fibrosis, periductal scarring due to prior trauma, excretory duct agenesis, ectopic sublingual glands or a tumor. Oral ranulas mostly originate from the secretions of the sublingual gland, they may also develop from the secretions of the submandibular gland duct or the minor salivary glands on the floor of the mouth. Cervical ranulas almost exclusively arise due to the mucus extravasation of the sublingual gland causes. The mucus escapes through openings or dehiscence in the underlying mylohyoid muscle.[4]

Earlier studies have shown increased levels of matrix metalloproteins, tumor necrosis factor-alpha, type IV collagenase, and plasminogen activators in mucoceles as compared to that of whole saliva. They causes increase in the accumulation of proteolytic enzymes which are responsible for the invasive character of extravasated mucus.[4]

Mucocele is a self-limiting mucous containing cyst of salivary glands commonly occurring in the oral cavity, with relatively rapid onset and with fluctuating size. The decrease in size may be attributed to either rupture of the lesion with subsequent mucin accumulation or re-absorption of saliva deposits may cause the lesion to reform.[10]

Mucus secretions can escape into the neck, through the mylohyoid muscle extending into the fascial tissue planes causing a diffuse swelling of the lateral or submental region of the neck. Rapid accumulation of mucus caused due to continuous secretions from the sublingual gland leads to a constantly expanding cervical mass. The sublingual gland, unlike the submandibular gland, is defined as a spontaneous secretor, capable of producing secretions without neural stimulation. Inflammatory reaction to these secretions results in the formation of granulation tissue and subsequent fibrosis that may result is the entrapment of the fluid and the sealing of the leak.[4]

The mucus retention cyst may also develop because of ductal obstruction; however, many of these lesions actually represent a distinct cystic entity of unknown cause. When ductal occlusion is involved, it is usually caused by a sialolith or an inspissated secretion that results in ductal dilatation and focal containment of the mucoid material.[4] Oral mucoceles, particularly Ranulas, may sometimes cause problems like discomfort, interference with speech, chewing, swallowing and swelling depending on the size and location of mucoceles.[10] Patient reported here had difficulty in mastication and swallowing alongwith the local discomfort.

Sialogram, ultrasonography, Magnetic resonance imaging (MRI), CT, and aspiration cytology can be helpful for diagnosis. Aspiration cytology will show mucin with muciphages and biochemical analysis will show increase in amylase and protein content.[3]

The microscopically, ranula is similar to that of a mucocele in other locations. The histopathologic spectrum includes the most common extravasation and rarely, retention variant. Histopathology ranges from acute inflammation intermingled with collection of mucin to mature lesions with scarce amounts of mucus and fibrosis of the stroma.[5] On microscopic examination, the mucocele shows an area of spilled mucin surrounded by a granulation tissue response. The inflammation usually includes numerous foamy histiocytes (macrophages). The

adjacent minor salivary glands often contain a chronic inflammatory cell infiltrate and dilated ducts. In some cases a ruptured salivary duct may be identified feeding into the area. The lesion may show hyperplastic parakeratinized stratified squamous surface epithelium. Presence of salivary gland tissue and sialomucin is diagnostic. Special stains like mucicarmine and alcian blue, are helpful in identifying mucin that is present freely in tissues or in the foamy macrophages.[7]

Surgery is the main stay for the management of ranulas. These include incision and drainage, excision of ranula, marsupialization, and marsupialization with packing or complete excision of the sublingual gland.[3] Cryosurgery, carbon dioxide lasers, electrocautery, intra-lesional injection of sclerosing agent OK-432 or steroid injection are also used for the treatment of mucoceles.[10] However, recurrence can occur and a new surgical intervention is necessary. Surgery may not be required if the ranula is small and asymptomatic. Marsupalization is another modality, where the intra-oral lesion is opened to the oral cavity with the aim of allowing the sublingual gland to re-establish connection with the oral cavity.[4]

CONCLUSION: Mucocele are mostly benign and self-limiting nature, primarily diagnosed based on clinical findings followed by definitive diagnosis based on the histopathological investigation.[1] The non-neoplastic diseases of salivary gland pose a diagnostic and therapeutic challenge to the clinician because of close resemblance of clinical presentation.

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