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RESEARCH ARTICLE

ADENOMATOID HYPERPLASIA OF LOWER LIP CLINICAL MIMICKER OF MUCOCELE CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT

Article History: Received 04th April, 2018 Received in revised form 12th May, 2018 Accepted 19th June, 2018 Published online 30th July, 2018 Adenomatoid hyperplasia (AH) is a rare lesion of the minor salivary glands characterized by localized swelling that mimics clinically with non neoplastic lesions (mucocele) as well as with neoplastic lesions of salivary glands. The lower lip is uncommon site of presentation. Histopathologically lesion comprised of variably sized lobular aggregates of normal appearing mucinous acini and duct with intervening thin fibroconnective tissue septae. Here, we report a rare case of Adenomatoid hyperplasia of lower lip in a 25 year old male patient, which mimics a mucocele clinically.

Key Words:

Adenomaatoid Hyperplasia, Miinor Salivary Gland,Lip, Mucocele.

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INTRODUCTION

Adenomatoid hyperplasia of minor salivary gland is a uncommon benign, non neoplastic asymptomatic enlargement of normal appearing salivary gland tissue in excess of that anticipated for that anatomic site. It was first described by Giansanti and Waldron in 1971 who reported intraoral mucinous minor salivary gland lesion presenting clinically as tumors (Giansanti, 1971). Very few case series have been reported in the literature and they have found hard and soft palate to be the most common sites (Barrett, 1995; Arafat *et al.*, 1981). However, other sites like retromolar area and labial mucosa have also been described (Buchner, 1991). The condition has been regarded as idiopathic with some role of chronic irritation and trauma in a few cases. It is difficult to clinically distinguish this lesion from other lesions like mucoretention cyst, sialadenitis, adenoma.

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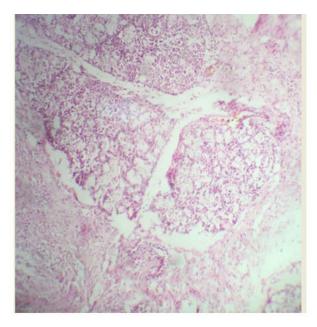
Here, we present a case of adenomatoid hyperplasia of minor salivary gland in lower lip which was clinically mistaken as mucocele and treated for it. The purpose of this report is to familiarize oral surgeons with this uncommon pathology of minor salivary glands, which should be differentiated from other non neoplastic lesions (mucocele) and as well from neoplastic lesions (adenoma) of minor salivary glands.

Case report: A 23 old male reported to our clinic with a asymptomatic, non tender, swelling in the labial mucosa of lower lip. The swelling was approximately 1.5cm with well defined margins, firm, sessile and normal appearing overlying mucosa with a bluish hue (fig1). The swelling was of six months duration with a history of trauma. Based on the clinical picture, history and site, the differential diagnosis of mucocele and adenoma was made. The lesion was excised under local anesthesia using sharp and blunt dissection and tissue was sent for histopathological examination. On gross examination it was labeled as a cyst 2cm in diameter with a smooth surface having wall thickness of 0.5cm. The tissue was fixed in 10% buffered formalin and was processed by Automatic tissue processor. The paraffin embedded tissue block were cut at 2-3 micron thickness and sections were stained haematoxylin and eosin.

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Fig. 1.A well defined submucosal nodular swelling of lower lip in a 25 year old male patient



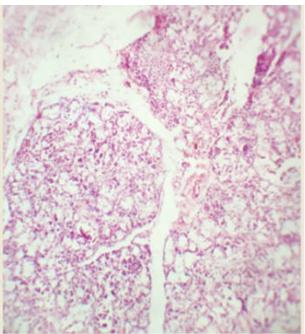


Fig 2 and 3. At lower power view shows lobular aggregates of normal appearing mucinous acini x10 H and E

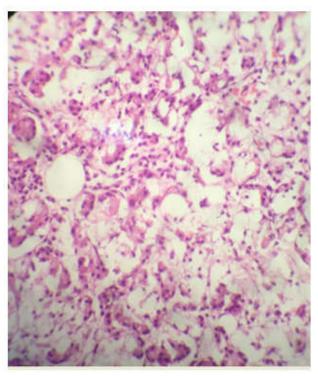


Fig. 4. High magnification shows distented mucinous acini surrounded by myoepithelial cells x40 Hand E

Under light microscopic examination lesion comprised of variably sized lobular aggregates of normal appearing mucinous acini and duct with intervening thin fibroconnective tissue septae (fig 2,3,4).The mucinous acini were slightly larger than normal, apperaed distended and were surrounded by myoepithelial cells. No mucous spillage was noted. No significant inflammatory infiltrate was seen in multiple sections. No dysplastic changes were noted. On the histopathology a final diagnosis of adenomatoid hyperplasia of minor salivary gland was made.

DISSCUSION

Adenomatoid hyperplasia (AH) is a rare lesion of the minor salivary glands characterized by localized swelling that mimics a neoplasm clinically but it is clearly distinguished histopathologically from salivary neoplasm as there is no abnormal cytologic and architectural features (Buchner et al., 1991). Hard and soft palate are the most common sites reported in case series by Bucher et al 1991, Barret et al in 1995 and Arafat et al in 1981 (Barrett et al., 1995; Arafat, 1981; Buchner et al., 1991). However, retromolar and labial mucosa sites too have been reported while upper and lower lip were the least common sites with AH (Buchner et al., 1991). It is seen with more incidence in males than females with a wide age range (9-79years) though fourth and fifth decade were the most common age for presentation. Most of the clinicians described it as asymtommatic, soft to firm, sessile, localized masses with normal appearing overlying mucosa though a few cases with pigmented mucosa are seen in the literature (Barrett, 1995; Neville, 2016). It has been reported with a size ranging from 0.5 to 3cms (Barrett, 1995; Buchner 1991; Sharma et al., 2011). The cause of this condition is uncertain yet though chronic irritation from sharp tooth, ill fitting denture and trauma has been suggested to have a role (Neville, 2016; Sharma, 2011). Endocrine disorders, nutritional deficiency, drugs or neuropathies implicated in asymptomatic swellings of major salivary glands have not been known to effect the minor salivary glands. This lesion cannot be categorized as Hamartoma as it most commonly presents in 4th to 6th decade and not during the developmental years (Barrett, 1995; Arafat, 1981). Our case was histopathologically diagnosed as adenomatoid hyperplasia of minor salivary gland although it was clinically diagnosed and treated as mucocele. The clinical appearance, site, bluish colouration and history of trauma in our case favoured the lesion clinically to be misdiagnosed as mucocele until proven histopathologically. Sometimes the presentation of the lesion on the palate has been described as "sheep in wolfs clothing" which can be mistaken as benign or malignant salivary gland tumor as mucoepidermoid carcinoma , soft tissue tumors as lympangioma, hemangioma and neurofibroma (Scully, 1992). The adenomatoid hyperplasia of minor salivary gland once completely excised have not yet reported to reoccur in the literature so far. To conclude, a good clinical and histopathological diagnosis is the key to report such cases in future literature. There has been no recurrence of this lesion after one year of follow

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