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Necrotizing Sialometaplasia-A Diagnostic Dilemma

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Abstract: ***Necrotizing sialometaplasia is an uncommon benign condition which masquerades as malignancy of minor salivary glands. The preservation of normal lobular architecture of the salivary gland with pronounced and extensive squamous metaplasia of ducts and acini of the salivary glands is the diagnostic feature of this condition [1]. We hereby present a case of 42 year old male with a submucosal swelling over hard palate which was diagnosed as minor salivary gland malignancy on fine needle aspiration cytology (FNAC) and turned out to be necrotizing sialometaplasia on post operative histo-pathological report.***

Keywords: necrotizing sialometaplasia, palatal sweeling, mucoepidemoid carcinoma of minor salivary glands

1. Introduction

Necrotizing sialometaplasia is a rare benign inflammatory condition which is locally aggressive but self limiting. It mimcs salivary gland malignancy on FNAC and hence may result in radical surgical resection of the disease than required, compromising the post operative quality of life of the patient. The most common presentation of necrotizing sialometaplasia is painful ulcerative lesion in the region of hard palate and rarely presents as a sub mucosal swelling. Here we present a case of 42 year old male with a smooth regular submucosal swelling in the region of hard palate which turned out to be a case of necrotizing sialometaplasia.

2. Case Report

A 42 year old male, farmer by occupation with no history of tobacco abuse or alcohol addiction presented with a complain of painless swelling over the palate giving a foreign body sensation while having food. On clinical examination, swelling was sub mucosal, smooth, regular, non tender situated in the posterior part of the hard palate on the right side. Overlying mucosa was normal with no discolouration or ulceration. Transillumination test was negative.

FNAC of the swelling was reported as having features suggestive of minor salivary gland malignancy. The type or grade of malignancy could not be commented upon. A CT paranasal sinus was done to rule out any bony erosion or defect if present at the base of the swelling. Coronal cuts of CT pns shows normal bony surface with no evidence of osteolysis. Hence, wide local excision of the swelling with 1 cm of margin was done by per-oral approach. The defect was repaired was using a local palatal rotation flap based on greater palatal artery of the same side. Post operatively, the wound was healthy and was completely mucosalised at 3 weeks. Post operative histo-pathology report revealed the specimen had preserved normal lobular architecture of salivary gland with evidences of lobular necrosis but no malignancy. There was presence of squamous metaplasia of acini and duct of salivary gland. Stromal infiltration with inflammatory cells especially neutrophils was present. There was no evidence of atypia, dysplasia or malignant transformation of the tissue. These all features are suggestive of necrotizing sialometapalsia.

3. Discussion

Necrotizing sialometaplasia is an uncommon, benign, locally aggressive but self limiting condition. This necrotizing inflammatory affliction of minor salivary glands was first recognized by Dr. John Cornyn and was described in detail by Albert Adams and Raymond Melrose in 1973 [1]. The age of presentation ranges from 1.5 to 83 years with most commonly presenting in the fourth decade [2]. Males are affected more commonly than females.

The cause of this condition is attributed to affliction of vascular supply to the salivary glands causing ishaemia which may result in necrosis. Traumatic injury, surgical procedure, heavy smoking, alcohol abuse, upper respiratory tract infections and associated lesions, local neurological deficit and allergies have been said to be contributory to this condition [2].

The most common site is the posterior part of hard palate followed by junction of the hard palate and the soft palate with two third lesions being unilateral. Other locations are soft palate, lip, retromolar trigone, tongue, mucobuccal fold, tonsillar fossa, parotid, sublingual, submandibular glands, nasal cavity, incisive canal, maxillary sinus and larynx [1].

The most common presentation is a painful but sharply circumscribed ulcer; frequently 1 to 3 cms in diameter. The ulcer borders are often erythematous and may be raised. In some instance, the mucosal surface is intact and the lesion is raised and fluctuant, giving false impression of an abcess[3]. Alves et al reported a case of necrotizing sialometaplasia presenting as non ulcerated nodule of hard palate [4]

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Anneroth and Hansen proposed 5 histological stages of pathogenesis of necrotizing sialometaplasia: infarction, sequestration, ulceration, repair and healing [5].

Carlson gave 5 charachterstic histological features of necrotizing siaalometaplasia[3] They are;

1. Pseudoepitheliomatous hyperplasia
2. Squamous metaplasia of ducts and acini
3. Preservation of lobular architecture
4. Lobular infarction with or without mucin spillage
5. Inflammation secondary to extravasation of mucin

The preservation of the normal lobular architechture is the key feature. The differential diagnoses of necrotizing sialometaplasia are mucoepidermoid carcinoma and squamous cell carcinoma [3]. Hence,in such cases immunohistochemistry can be helpful when the histological picture is overlapping. The incorporation of an antibody panel including myoepithelial markers (smooth muscle antibody, p63, calponin), basement membrane markers (laminin, collagen type IV), E-cadherin, and various cytokeratins (CK5, CK6, CK7, CAM 5.2) has been suggested [6, 7]. Both MIB-1 and p53 can stain positively in either benign or reactive squamous epithelium but labelling is generally more intense and is increased in malignancy but alone,neither is diagnostic[3].

The disease is self limiting and the ulcers, if present, heal within few weeks after presenation. The nodular swelling can be excised if not remitting after observation for atleast two months.

4. Conclusion

Necrotising sialometaplasia is a self limiting benign but locally aggressive disease. It is often confused with mucoepidermoid carcinoma of minor salivary glands on FNAC but preserved normal lobular architecture of salivary gland is the characteristic distinguishing feature. Hence this differential should always be considered during management of palatal lesion to prevent unnecessary radical approach and to give a better quality of life to the patient.

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Figure 1: CT PNS showing a smooth regular swelling on the hard palate

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