

Chronic Sclerosing Sialadenitis of the Minor Salivary Glands

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Abstract

Chronic sclerosing sialadenitis (Kuttner's tumor) is a chronic inflammatory condition of the salivary glands; commonly the submandibular salivary gland that cannot be distinguished from a true salivary gland neoplasm. This is a report of chronic sclerosing sialadenitis involving a minor salivary gland that occurred as an isolated mass in the region of the cheek with unusual clinical features.

Keywords: Chronic Salivary Sialadenitis; Minor Salivary Glands

Introduction

Chronic sclerosing sialadenitis (CSS) is an inflammatory disease of the salivary glands, that clinically appears as a hard swelling commonly the submandibular gland [1] which cannot be distinguished from neoplasia. In 1896, Kuttner [2] was the first to describe this disease in the submandibular gland of middle-aged adults, which was not associated with autoimmune or other fibrosing diseases. This lesion always raises a strong suspicion of a neoplasm in the mind of the clinician due to its manifestation as a hard mass. We report a case of chronic sclerosing sialadenitis of the minor salivary glands located in the right cheek, which was asymptomatic, closely mimicking a neoplasm with an uncertain etiology. There was no association with any other major salivary glands preceding or following the excision of this lesion.

Case Report

An 80-year old male patient reported to our department with a chief complaint of an asymptomatic hardness of the skin of the right cheek since last 3 months. The lesion (Figure 1) was 3.5 cm in diameter, which was firm, painless, well circumscribed and adherent to the underlying mucosa as well as the skin. The patient had no relevant systemic debilitating diseases. Also the facial nerve functions were within normal limits with no lymphadenopathy. An ultrasonography (USG) was advised which revealed an irregular hypoechoic solid nature lesion with indistinct margins which was relatively avascular prompting it to be a neoplastic lesion. The nature of the lesion was further confirmed by fine needle aspiration cytology (FNAC) that revealed an inflammatory infiltrate of polymorphs, a few sheets of ductal epithelial cells and mucin in the background proclaiming chronic sialadenitis but at the same time not ruling out low grade mucoepidermoid carcinoma.



Figure 1: Clinical Picture.

In view of the asymptomatic state and absence of lymphadenopathy, the lesion was excised under general anesthesia. The excision site was then reconstructed using buccal pad of fat (Figure 2) thereby preventing any dead space, with the skin closure done using 6-0 prolene sutures. The histopathology later revealed a fibrocellular connective tissue, presence of lymphoplasmocytic infiltration (Figure 3) with sclerosis (Figure 4) in several places and areas of acinar destruction that confirmed a diagnosis of CSS of the minor salivary glands.



Figure 2: Surgical Excision and Reconstruction.

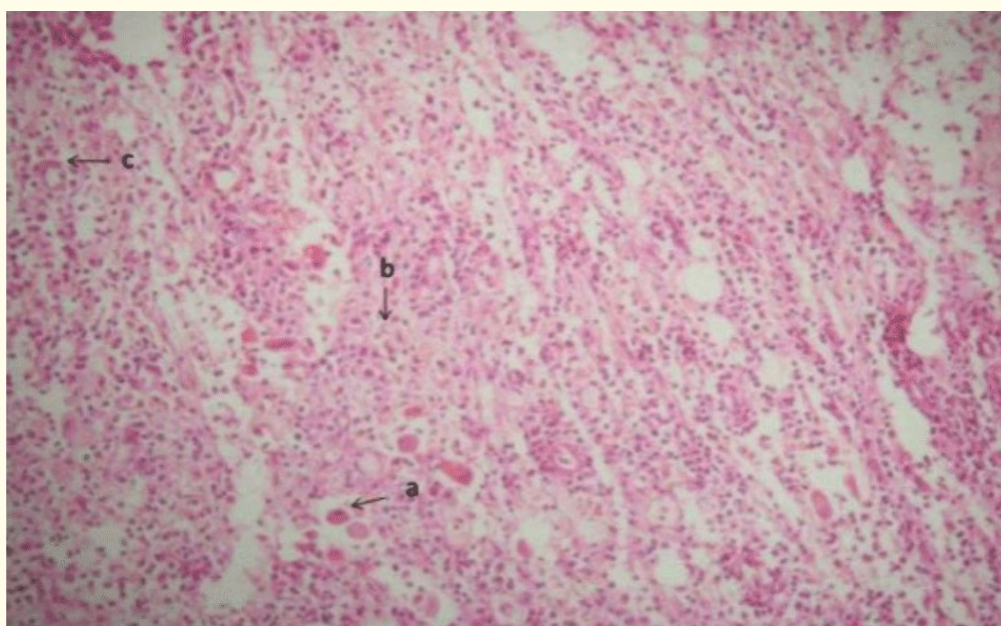


Figure 3: Histopathology (40 X Magnification).

a: Areas of destructed acinar cells; *b:* Connective tissue with lymphoplasmocytic infiltration; *c:* Blood vessels.

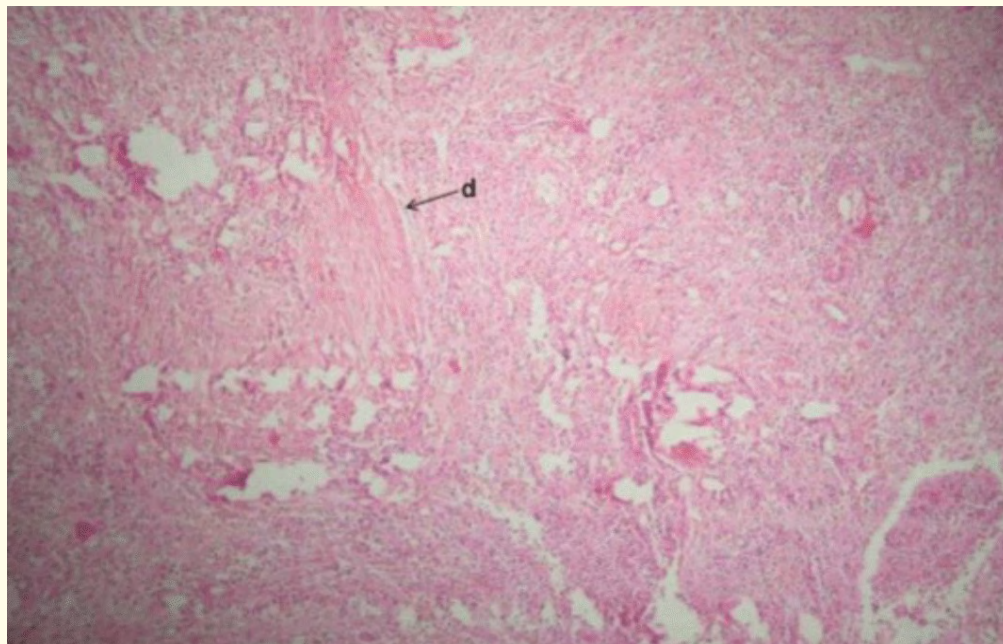


Figure 4: Histopathology (20 X Magnification).
d: Sclerotic fibrous connective tissue.

Discussion

CSS has been documented as a unique clinicopathologic entity in the classification of tumor like lesions of the salivary glands. This disease is symptomatic or asymptomatic, with clinical symptoms being rendered as recurrent pain and swelling in the submandibular glands occurring usually during mealtimes [3]. In our case there was no pain or any swelling associated with the lesion either during mealtimes or later and the rigidity of the lesion remained constant at all times. The salivary flow as per the age of the patient was normal and he had no difficulty in deglutition. As per the description of the same lesion given by Williams., *et al.* [4], their patient had a resolution of the swelling during a prolonged fast. This does not comply with our patient where absolutely no resolution was observed. Intermittent swelling with resolution was observed over weeks and months in immunologic sialadenitis [5].

In 1977, Seifert and Donath described four degrees of severity of inflammation in CSS, which could be distinguished histologically [6]. Our case exhibited a stage 3 as per this histopathological classification, i.e. chronic sclerosing sialadenitis with salivary gland sclerosis. This confirms the existence of numerous clinical variants with differing histopathology.

Sialolithiasis is the most common etiologic factor associated with chronic sclerosing sialadenitis, occurring in 50% to 83% of cases, with the sialoliths most frequently being extraglandular [3]. Sialoliths could cause local inflammation thereby causing infection ultimately leading to CSS. This postulation as an etiologic factor although still remains ambiguous [7]. Also immune reactions of the duct systems, lymphocyte inflammation and disorders of secretion have been proposed as likely causes [8]. Tsuneyama., *et al.* [9] have reported an association of CSS with autoimmune diseases, wherein there is replacement of glandular tissue by lymphoid and fibrous connective tissue [10]. With respect to our case the etiology remains questionable.

As is our case with a bizarre location of the lesion, existing skin induration and a complete absence of any associated salivary gland dysfunction, a clinical diagnosis of “any” salivary gland disease could have rarely been made. With the advent of an array of a variety of presentations in literature and the facilitation of newer diagnostic aids, the clinician is aptly benefitted to correctly diagnose and devise the best treatment plan for such varying lesions.

Conclusion

As is our case with a bizarre location of the lesion, existing skin induration and a complete absence of any associated salivary gland dysfunction, a clinical diagnosis of “any” salivary gland disease could have rarely been made. With the advent of an array of a variety of presentations in literature and the facilitation of newer diagnostic aids, the clinician is aptly benefitted to correctly diagnose and devise the best treatment plan for such varying lesions’.

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