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Case Report

Unilateral chronic sclerosing sialadenitis of obstructive etiology: A case report

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ABSRACT:
Sialadenitis is a non-neoplastic inflammatory condition of salivary glands, which may be acute or chronic. There are various causes of sialadenitis however sialolith has been reported as the most common etiologic factor. Sialolith is the formation of salivary calculi as a result of deposition of inorganic salts around an organic nidus such as thick mucus, exfoliated epithelial cells, salivary proteins and foreign bodies. This causes the blockage of salivary duct, with reduction or stasis of the salivary flow, followed by retrograde bacterial invasion, leading to acute or chronic infection in the gland. Here we report a case of chronic submandibular sialadenitis associated with multiple sialoliths in a middle aged male patient. Diagnosis was confirmed using clinical, radiographic and histological findings.
Keywords: Sclerosing Sialadenitis, sialolith, Submandibular Gland

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INTRODUCTION

Chronic sclerosing sialadenitis is the most common non-neoplastic disease of salivary glands and was first described by Kuttner in 1896.[1]In WHO classification it is classified as tumor like condition of salivary glands. It is important to distinguish the lesion from various neoplastic as well as non-neoplastic diseases of salivary glands such as lymphomas and other inflammatory conditions.

In 50 to 83% of cases the condition is associated with sialolithiasis, rest of the cases had disorders of secretion, lymphocytic inflammation and immunological etiology.[2] More commonly seen in male patients than females with a ratio of 2:1.[3]

Clinical findings in these cases may or may not be symptomatic. Lesion often presentsas palpably firm swelling, with varying degree of tenderness in the associated gland area. Chiefly affecting the submandibular duct system due to long tortuous course of its duct in which inspissated saliva travels against gravity, which makes it more prone to stasis and retrograde bacterial invasion. In the sub-mandibular duct, stones are often found at the duct's bend around the posterior edge of mylohyoid muscle.[4]Any association with altered serum calcium levels or renal stones has not been reported so far. In addition to clinical findings various other means of investigations such as OPG, CT, USG, FNAC and histopathological examination are also useful to conclude the diagnosis.

Conventional radiographs show radio-opaque calculi, with an accuracy of 92%.[5]Occlusal radiographs are of help, only if the stones are located in anterior portion of the duct. To identify the stones within the glands, a CT scan is recommended as the most reliable investigation, which eliminates the need for sialography. Sialoliths located more posteriorly, within the glandular parenchyma or at the posterior border of mylohyoid are often confused with the calcifications from other sources, such as calcified lymph nodes, phleboliths, tonsoliths or calcifications from carotid bifurcation.[4]

Usually ultrasonogram of this lesion shows diffuse involvement of salivary gland with multiple small hypoechoic foci with homogenous background.[6]The role of USG in calculi detection shows 94% sensitivity and 100% specificity, however, this investigation tool seems to be less precise in differentiating a cluster of stones from a single large stone.[7]

In cases of sialolith induced chronic sialadenitis, histopathological examination reveals dilated duct with calculi, chronic inflammation, acinar atrophy and fibrosis. While in sialadenosis, parenchymal atrophy and compensatory adiposis is found.[8]

Seifert and Donath had proposed 4 progressive histopathological stages of this lesion depending on the degree of inflammation [9]

a) Stage 1 – Focal Sialadenitis
b) Stage 2 – Diffuse lymphocytic sialadenitis with salivary gland fibrosis.
c) Stage 3 – Chronic sclerosing sialadinitis with salivary gland sclerosis.
d) Stage 4 - Chronic progressive sialadinitis with salivary gland cirrhosis.

Later in 2001 Francis Marchal *et al* introduced another classification based on degree of atrophy, fibrosis and inflammation [10]

1. Grade A – No atrophy and no fibrosis (score 0), and absent or moderate inflammation (Score 0 or 1).
2. Grade B – moderate atrophy (score 1), and/or fibrosis (score 1), with any stage of inflammation (0 to 2)
3. Grade C – important atrophy and/or fibrosis (score 2) with any stage of inflammation (score 0 to 2)

Our aim of reporting this case was to correlate the clinical findings of submandibular gland swelling caused by sialolith with histological features.

CASE REPORT

A 53 years old male patient reported to the OPD of AMC Dental College and Hospital, Ahmedabad, with the chief complain of firm swelling below the lower jaw on right side, since two and half months. Earlier the swelling was small pea sized, and had slightly increased in size over a period of 15 days. Patient reported mild occasional pain in swelling, but had no history of fever or any discharge from the area. No history of increase in size of the swelling on seeing or eating food. Past medical, surgical, allergic history was non-significant, however, he got his 46, 47 extracted under LA, 20 years back, with no complications. No relevant family history. Patient was a habitual bidi smoker since 20 years and smoked 4 to 5 bidis per day. All vitals were in normal range. On inspection, facial asymmetry was evident with a single,2 x 3 cm, lobulated swelling present on the right side submandibular region, with normal overlying skin and no bleeding, pus discharge or sinus tract. On palpation the swelling was non-tender, firm, non-compressible and mobile.Patient was advised further diagnostic workup. OPG findings revealed a single, small ill defined radio opaque mass of size approximately 6x8mm at right side of mandibular body below mandibular canal, suggestive of sialolithiasis. No stone was noticed in occlusal radiograph. CT scan of right submandibular region suggested hyper dense and enlarged gland with heterogeneous post contrast enhancement;presence of few hyper dense foci in the path of submandibular duct-largest being 7x55mm-thus, a possibility of sialolithiasis was put forward. Lymph node enlargement of levels Ia, II,IIb,III, IV was also noticed, thus a diagnosis of sialadenitis with sialolithiasis on right side with cervical lymphadenopathy was given. USG report revealed internal specks of calcifications of size approximately 35x22mm in submandibular region with altered echo pattern of right submandibular gland and hypoechoic level II cervical lymph node.FNAC findings suggested inflammatory salivary gland lesion. Right Submandibular salivary gland was excised with sialoliths, under GA. Excised salivary gland along with involved duct and stone was submitted to the oral pathology department for histopathologic diagnosis. Formalin fixed paraffin embedded tissue revealed preserved lobular architecture of salivary gland with dense septal fibrosis. Periductal fibrosis and hyalinization with mucous and squamous metaplasia in duct was evident at some places, along with diffuse infiltration of lymphocytes and lymphoid aggregates forming germinal centers. Acinar atrophy was also seen, consistent with the diagnosis of chronic sclerosing sialadenitis. Patient was followed up for salivary gland function test; in which it was found out that left salivary flow rate was normal, while salivary flow rate on right side of lingual frenum was reduced. No adverse outcomes of gland extirpation were reported by the patient on 4 year follow up.

Fig 1 – OPG shows radioopaque calculus near right mandibular angle region.

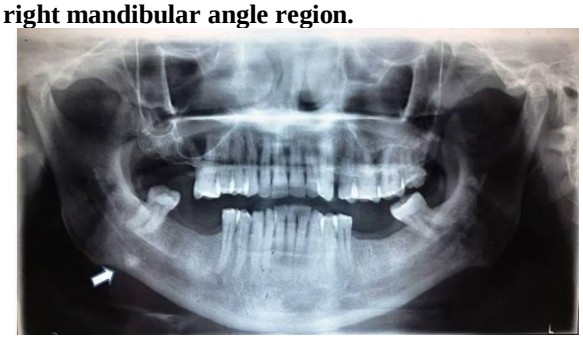


Fig 2 (5x) – lower power view shows preserved lobular architecture, stromal fibrosis (A) and lymphoid aggregated with germinal centers (B).

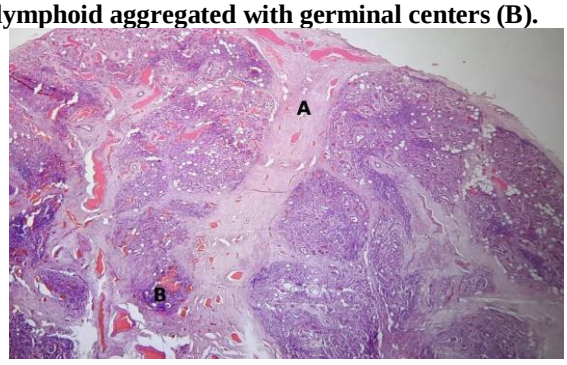


Fig 3 (10x) – 10 x view shows preserved architecture of lobules however there is extensive replacement of parenchyma by fibrosis and lymphocytic infiltration (A).

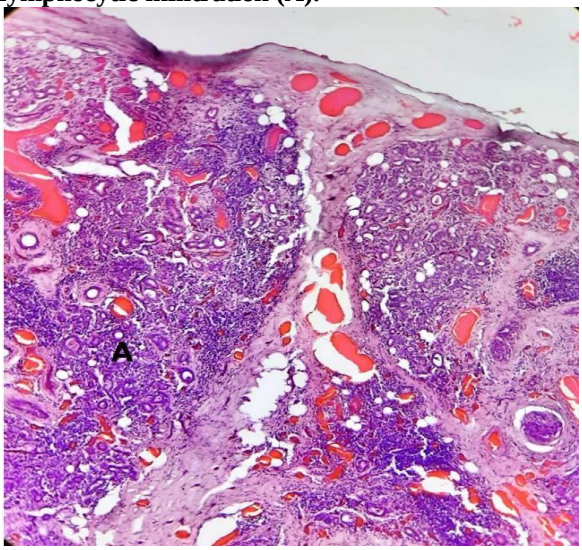
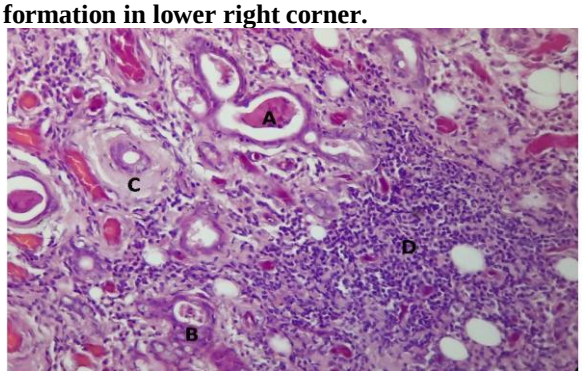


Fig 4 (40x) – 40x view shows dilated duct with eosinophilic secretions(A), ductal squamous metaplasia (B), periductal fibrosis and hyalinization (C) with lymphocytic follicle (D) formation in lower right corner.



DISCUSSION

Chronic Sclerosing Sialadenitis (CSS) is non-neoplastic disease, in which symptoms varies from intermittent painless swelling, moderate discomfort to severe pain, that may or maynot be associated with the food intake.[6] Patient discussed here had similar clinical findings.The lesion being a hard massmimics clinically as a malignant salivary gland neoplasm, so it is important to rule out salivary gland lymphomas, mainly sclerotic lymphoma (nodular form of Hodgkin's disease) and MALT lymphomas. [11]

Many theories regarding etiopathogenesis of CSS have been proposed, where the disease is thought to be related to autoimmune response in the form of CD3, CD4 and CD8 positive T-Cell predominance;[12] and/or its relation with elevated levels of IgG4.[1] Among healthy population, IgG4 is the smallest component of IgG subclass, whose biological function is still unclear. Sclerosing diseases of many organs such as pancreas, biliary tree, liver, gall bladder, lacrimal gland, salivary glands, kidney, lung pleura and lymph nodes, have been related to more than 50% of IgG4 positivity in IgG subclass.[9]In our case multiple stones were detected on radiographic examination so, the diagnosis was more inclined towards obstructive etiology instead of immunologic.

Ultrasonography has limitations in differential diagnosis of CSS from malignancies because both have almost similar findings such as diffuse involvement of the gland with multiple small hypoechoic foci scattered on a heterogeneous background. In addition in our case specks of calcifications were noted, thus verification with further investigations was mandatory.

CT pattern can identify gland suspicious for a concomitant tumor.[4]Usually inflammation and fibrosis of glands show homogenous pattern. While tumors show heterogeneous pattern, similar to the CT findings in our case, thus for confirmatory diagnosis histopathological examination was more reliable tool.

Based on histological examination, primary diagnosis of CSS was made, which was reinforced by the findings suggested by*Chiang AK et al*,[13] that, malignant tumors like MALT lymphomas are not associated with CSS, due to reduced B cell response in it.Other immunological diseases like Sjogrens syndrome and Myoepithelial Sialadenitis (MESA) were ruled as lymphocytic infiltration was not typical monocytoid B cell type and fibrosis is not a feature of these diseases in major salivary glands.[2]

As per the classification of histological stages by Seifert and Donath in 1977, our case fits in the stage 2, while according to the classification put forward by Francis Marchal*et al* in the year 2001 findings were consistent with those described in grade B.

Inflammation is seen in initial phases of the disease which is a partially reversible process. If inflammation persists, over time fibrosis and atrophy develop and the gland becomes non-functional, thus later considered better predictors of the restoration of glandular function post disease.[4,10]Considering the advanced stage of disease in our case, complete extirpation of the gland was suggested.

CONCLUSION

CSS is a rare and under-reported non-neoplastic salivary gland lesion which mimics salivary gland neoplasms, thus it becomes a perplexing situation for clinician. So highlights the key role of pathologists, in treatment planning.

It seems further research is needed to understand the etiopathogenesis of this disease, as it is still unclear whether sialadenitis follows sialolith formation or vice versa.

Autoimmune etiology should be ruled out at first place to prevent involvement and progression of the disease in other organs prone for the same.

In case of early diagnosis the lesion can be treated by non-invasive methods, with favorable outcome and good long term prognosis.

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CONFLICT OF INTEREST
There are no conflicts of interest.

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