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RESEARCH ARTICLE

SIALOGRAPHY FOR SJOGREN'S SYNDROME: AN IMPORTANT DIAGNOSTIC TOOL

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Abstract

Sjogren's syndrome (SS) is a chronic autoimmune disorder that affects the exocrine glands. Primary Sjogren's syndrome is associated with dry mouth; dry eyes, and is frequently found in middle middle-aged females in their fourth or fifth decade. This article presents an atypical case of Sjogren's syndrome in a 21-year-old woman who complained of bilateral parotid swelling and diagnosis was made with the help of parotid sialography. Though the confirmatory diagnosis was achieved with the help of serological and histopathological examination, an oral medicine specialist can play an important role in the detection of this disease.

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Introduction:-

Sjogren's syndrome (SS) is named after Swedish ophthalmologist Henrik Sjogren in 1933.¹ Sjogren's disease can be further classified into two types; first type is Primary Sjogren's syndrome which is associated with dry and mouth (sicca complex) and extra glandular symptoms without connective tissue disorder. The second type is Secondary Sjogren's syndrome which includes Sicca complex with autoimmune disorder like rheumatoid arthritis, systemic lupus erythematosus. Most people are more than 40 years old at time of diagnosis, with female: male ratio of 9:1.² However, the youngest reported case of SS was a two-year-old patient.³ This article presents a rare case of primary Sjogren's syndrome in a young female with the clinical presentation of bilateral parotid gland swelling only, and as well as effectiveness of sialography as a diagnostic tool.

Case History:

A 21-year-old female reported with the complaint of recurrent swelling of bilateral parotid glands since last one year. Patient was alright prior to one year, when she first noticed swelling in the region of left parotid gland associated with fever, pain and swelling which subsided with oral antibiotics and analgesic course. Afterwards, there was recurrent course of non-tender swelling with no report of fever, foul smell, or change in taste and similar pattern of involvement of left and right parotid gland as mentioned above in past three months. The swelling used to subside completely with a course of oral antibiotics. However, the patient reported with the complaint of bilateral parotid swelling which had not subsided with the intake of oral antibiotics this time. No history of oral dryness, joint pain, ocular dryness, photosensitivity, facial rash, alopecia, mouth ulcers, foreign body sensations were present. Medical and family history was non-contributory.

On extra oral examination, swelling was soft in consistency, non-tender seen bilaterally in pre auricular region extending up to post auricular area [Fig.1]. The swelling was single with diffuse borders, no evidence of erythema, draining sinus or ulceration on overlying skin.

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Intra-orally, no history of oral dryness was reported. The salivary ducts were patent on cannulation. Based on history and examination, provisional diagnosis of recurrent parotitis secondary to calculi or chronic infection and differential diagnosis of Sjogren's syndrome, sarcoidosis, and benign salivary gland neoplasm were given.



Figure 1:- Extraoral view showing swelling over the right and left lower two two-third region of the face.

The investigations advised were Orthopantomogram (OPG), Ultrasonography (USG) of parotid and submandibular glands, sialography, chest X-ray, autoantibody test, antinuclear antibody test, and lower labial mucosal biopsy. Chest X-ray was normal. Sialography of the right and left parotid gland was performed. The radiograph OPG revealed presence of minute amount of radio-opaque contrast agent in the parotid gland in foci, which was suggestive of punctate sialectasia (approximately 1 - 2 mm). The presence of widespread dots and blebs of contrast medium within entire gland bilaterally was suggestive of fruit laden branchless tree appearance, leading to a provisional diagnosis of Sjogren's syndrome [Fig. 2,3].

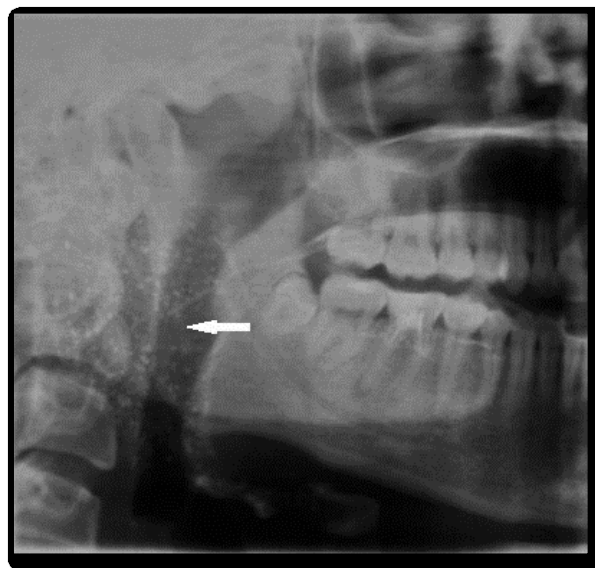


Figure 2:- Orthopantomogram of right side showing multiple globular collection of the contrast on sialography in the parotid region.

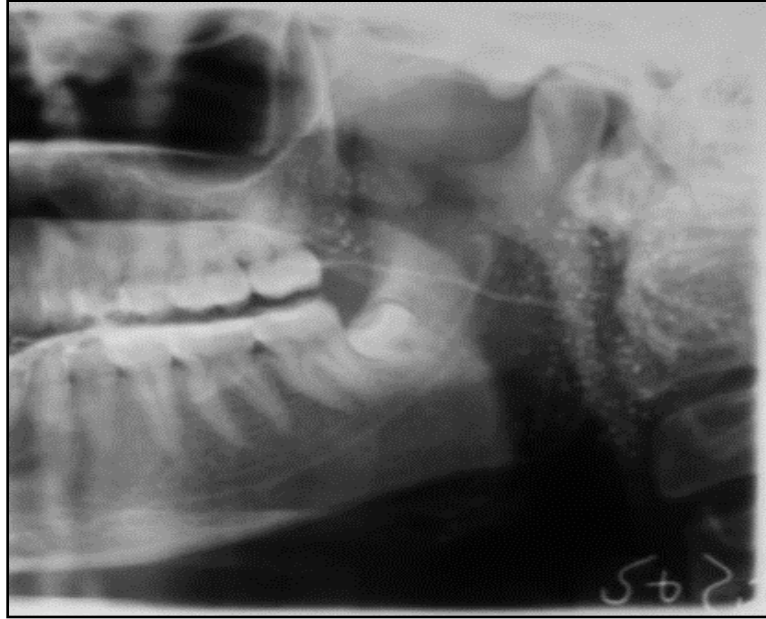


Figure 3:- Orthopantomogram of left side showing multiple globular collection of the contrast on sialography in the parotid region.

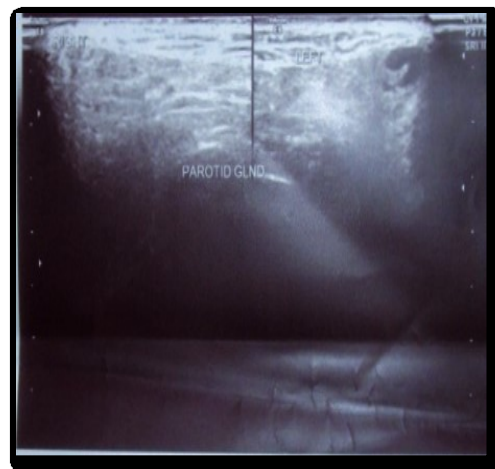
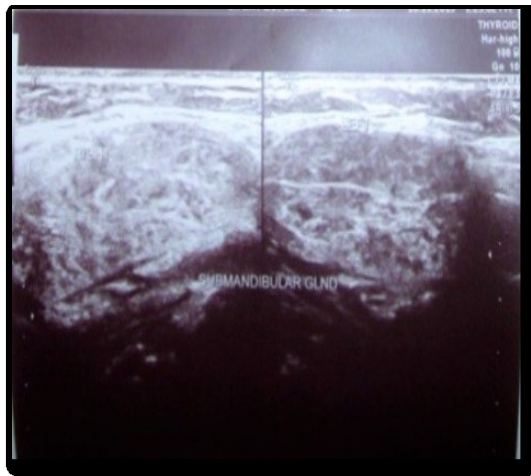


Figure 4:- Ultrasound image of Parotid gland **Figure 5:-** Ultrasound image of Submandibular gland.

Neck USG revealed that both parotid and submandibular glands were mildly bulky and showed diffusely altered parenchymal echotexture with small hypoechoic lesions scattered throughout the parenchyma [Fig.4,5]. Small subcentimetric intraparotid lymph nodes were noted. Antinuclear antibody test by immunofluorescence revealed positive grade ++ (titre 1:320), positive SSA-Ro [value - - 191.72 RU/ml] and positive SSA-La autoantibody [value- - 116.28 RU/ml] levels. Incisional biopsy from the lower labial mucosa revealed presence of focal aggregates of mononuclear inflammatory cells evident in peri-acinar and peri-ductal distribution [Fig.6]. A confirmatory diagnosis of primary Sjogren's syndrome was given. Patient was advised short term steroid course, adequate water intake, in between sips of lemon juice, multivitamin supplement, and artificial saliva for oral symptoms. On follow-up, significant reduction in the symptoms was seen. Unfortunately, the patient did not report back and was lost to future follow-up.

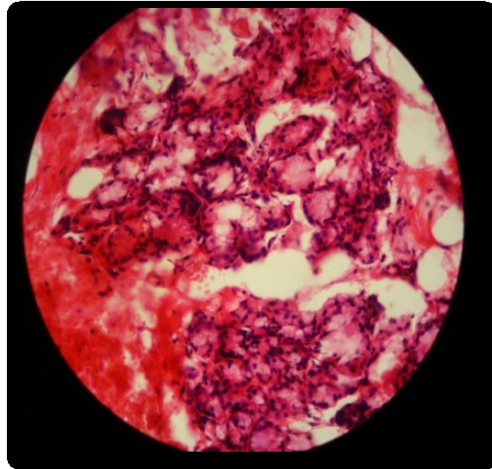


Figure 6:- Histological examination of the sub-labial salivary gland tissue showing numerous salivary gland acini and a dense chronic inflammatory cell infiltrate (40×X).

Discussion:-

Dry mouth (xerostomia) and bilateral parotid swelling, as in our case, may be a symptom of many other conditions such as patients on medications, multiple sclerosis, Alzheimer's disease, and sarcoidosis. To arrive at a diagnosis of Sjogren's syndrome, the revised criteria proposed in 2002 by the American European Consensus Group criteria (AECC) should be evaluated. The following criteria should be met for diagnosis of Sjögren's syndrome:

Ocular Symptoms (at least one):

1. Dry eyes symptoms for 3 months at least
2. A foreign body sensation in the eyes
3. Artificial tears use for 3 or more times in a day

Oral Symptoms (at least one):

1. Dry mouth symptoms for 3 months at least
2. Recurrent or persistently swollen salivary glands
3. Need for liquids to swallow dry foods

Ocular Signs (at least one):

1. Abnormal Schirmer's test, (without anesthesia; ≤ 5 mm/5 minutes)
2. Eye surface staining with vital dye is positive

Histopathology:

1. Lip biopsy showing focal lymphocytic sialadenitis (focus score ≥ 1 per 4 mm²)

Oral Signs (at least one):

1. Un-stimulated whole salivary flow (≤ 1.5 mL in 15 minutes)
2. Abnormal parotid sialography
3. Abnormal salivary scintigraphy

Autoantibodies (at least one):

1. Anti-SSA (Ro) or Anti-SSB (La), or both
2. For primary Sjögren's syndrome diagnosis:
3. Out of 6 any 4 criteria should be met and must include either item IV (Histopathology) or VI (Autoantibodies)
4. Out of 4 any 3 objective criteria (III, IV, V, VI) must be met.

For secondary Sjögren's syndrome diagnosis:

The presence of one symptom (I or II) plus 2 of 3 objective criteria (III, IV, and V) may be indicative of secondary Sjogren's syndrome. In present case, four of the six criteria including IV and VI were satisfied, thus confirming the

diagnosis of primary Sjogren's syndrome. This syndrome requires management by oral and general medicine department. Treatment of oral symptoms involves oral hygiene maintenance, fluoride application, restoration of dental caries, use of artificial saliva, abundant water intake, multivitamins, lemon drops and muscuranic agent's helps to increase saliva secretion.^{4,5,6}

Sometimes patients presenting with only symptom of salivary gland swelling can be difficult to diagnose, in such cases, sialography can be a reliable diagnostic tool for diagnosis followed by histo-pathological examination and serological tests for confirmation. Patients should be informed about progress of disease, its various signs, symptoms and methods to treat these symptoms.

Source of Support:

None.

Conflicts of Interest:

There are no conflicts of interest.

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