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Management of Sjogren's Syndrome induced Xerostomia with Artificial Saliva- A Case Report

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Abstract

Sjogren's syndrome (SS) commonly known as "Sicca Syndrome" is a chronic, systemic autoimmune disease characterized by keratoconjunctivitis and xerostomia resulting from lymphocytic infiltration of the lacrimal glands and salivary glands resulting in glandular dysfunction. It occurs worldwide mainly in middle aged women with a female predisposition and rarely in children. It may exist as primary or secondary SS. Oral manifestations of SS help in early diagnosis and management of the disease. We present a case of secondary Sjogren's syndrome and management of xerostomia induced by Sjogren's syndrome with Artificial Saliva.

Keywords: Sjogren's syndrome, Xerostomia, Sicca syndrome, Artificial Saliva

Introduction

Sjogren's syndrome is a chronic, systemic autoimmune disease characterized by lymphocytic infiltration of exocrine glands. It mainly involves lacrimal and salivary glands leading to two typical features of the disease: Keratoconjunctivitis sicca and xerostomia.[1] Sjogren's syndrome occurs in two forms: Primary Sjogren's syndrome, when its clinical manifestations are seen alone and Secondary Sjogren's Syndrome, when associated with another autoimmune disease, most commonly Rheumatoid arthritis(20–32%), Systemic lupus erythematosus (15–36%), as well as limited and progressive systemic sclerosis (11–24%).[2] The prevalence of SS is estimated to be approximately 3% in subjects 50 years or older, with a female to male ratio of 9:1.[3] The patients suffering from Sjogren's Syndrome visit the dental clinic for the discomfort associated with xerostomia. Till date many modalities have been tried for the treatment of Xerostomia

induced by Sjogren's Syndrome and other systemic conditions. Here we present a case report wherein we managed Xerostomia induced by Sjogren's syndrome with Artificial Saliva after weighing the pros and cons of available treatment modalities.

Case Report

A 38-year-old female patient reported with the chief complaint of Dry mouth and Dry eyes since 1 year. Patient also complained of burning sensation in the oral cavity since last 6 -7 months. Her past medical history revealed that she had Joint pain since 2 years which was severe in early mornings. Also, patient gave history of Bronchial asthma since last 4-5 years for which she has stopped taking treatment. There was presence of pain, stiffness and swelling over joints, especially ankles, wrists, fingers & knees (fig.1). Patient also had difficulty in carrying out household work. Pain was so severe that patient couldn't make a fist. On general examination swelling was seen over the joints of hands. Mobility of fingers and wrists was reduced. Dryness of nasal & esophageal mucosa with difficulty in swallowing hard, sticky food stuffs was seen. There was Dryness, itching of eyes since a year along with foreign body sensation within eyes, more pronounced in the left eye. Extra-oral examination revealed dry, cracked lips (fig.2). There was presence of diffuse swelling on the right and left side parotid region, soft and non-tender on palpation (fig.3).

Intraoral examination of the soft tissue revealed that the nature of saliva was scanty, thick / pasty, ropy with no foul smell (fig.4). There was Dryness of all mucosal surfaces with minimal salivary pool in the floor of the mouth (fig.5). Mucosa was pale. Tongue blade sign and lipstick sign were positive. Patient had difficulty in speech, mastication and deglutition. Sialometry showed Unstimulated salivary flow rate less than 0.1 ml/min and Stimulated salivary flow rate- 2 ml/min Patient had multiple caries in posterior region. Depending on these findings a provisional diagnosis of Sjogren's Syndrome was established and further investigations were carried out.

Ultrasonography of Parotid gland (right and left) was done. Right parotid measured 3.6x1.2x2.1 cm and Left parotid measured 3.9x1.2x1.4 cm. Both parotid glands were mildly increased in size, and heterogenous in architecture. Well defined reactive lymph node measuring 10.4 x 5.1mm was noted in left parotid gland. Findings were suggestive of parotitis. Hematological and serological tests were done. RA factor was 16.4 IU/ml. It was found to be weak positive. Also, she had weakly positive antinuclear antibodies (ANAs). Sialography of Parotid glands revealed narrowing of ducts and ductules along with reduced salivary flow. Schirmer's test showed tear deficiency which is <5 mm wetting of the paper after 5 minutes.

Histological examination revealed aggregates of lymphocytic infiltration consisting of T lymphocytes around ducts. Proliferation of ductal epithelium was also seen some of which were obliterating the lumen (fig 6). The histological features confirmed the diagnosis of Sjogren's syndrome.

The final diagnosis of Secondary Sjogren's Syndrome was made as she had Rheumatoid Arthritis. Patient was reassured and oral hygiene instructions and dietary advice were given. Restorative treatment of carious teeth was started. Patient was advised frequent sipping of water, rinsing of mouth after meals or snacking and avoidance of tart, hard, sticky and sweet foodstuffs. As Pilocarpine is contraindicated in patients with SS with uncontrolled asthma, our patient was symptomatically treated by Artificial saliva. Patient was given "Wet mouth", liberal application was advised before meals thrice a day for 4 weeks. Improvement was seen after 6 days. Dryness and

burning sensation were relieved and patient was able to chew on hard food. An ophthalmology referral was done for further management of ocular symptoms.

Discussion

Sjogren's syndrome is a chronic, auto immune disease characterized by dry mouth, dry eyes and parotid gland swelling. Pathophysiology of SS is chronic immune system activation leading to humoral and cellular autoimmune reaction.[4] Destruction of the salivary and lacrimal glands due to infiltration and proliferation of CD4+ T cells, B cells and plasma cells leads to atrophy of acini resulting in xerostomia and keratoconjunctivitis sicca.[4] the exact mechanism of activation of autoimmune reaction is uncertain. Both T and B cells are found to be involved and B cell hyperactivity is uttered through hypergammaglobulinemia and circulating auto antibodies.[5] There are organ specific and non organ specific auto antibodies involved which contribute to tissue dysfunction. Organ specific auto antibodies comprise of antibodies to cellular antigens of the salivary ducts, the thyroid gland, the gastric mucosa, erythrocytes, the pancreas, the prostate and nerve cells where as non-organ specific antibodies include rheumatoid factors, antinuclear antibodies and antibodies to the small RNA protein complexes SS-A (Ro) and SS-B (La).[4]

The diagnosis is made in patients having experienced sicca symptoms that is presence of dry eyes (xeropthalmia) and dry mouth (xerostomia) for at least 3 months, these are based on further function tests (e.g. the Schirmer's test) or serological (Anti-Ro/SSA) and histological examinations (labial salivary gland biopsy). Among these investigations, anti-Ro/SSA antibodies and an abnormal labial salivary gland biopsy have the highest specificity; consequently, they are the criteria with the highest values.[2] The histopathological findings are focal periductal localized lymphocytic infiltrates in the exocrine glandular tissue along with otherwise intact acinar units. These findings are pathognomonic for Sjogren's syndrome. These infiltrations mostly consist of CD4+ T cells, with some additional CD8+ T cells and CD19+ B cells, plasma cells and dendritic cells.[2]

Clinical manifestations of patient with SS include xerostomia, xerophthalmia dryness of the skin, respiratory tract and the vagina. In addition to this there can be symptoms and signs of other connective tissue autoimmune diseases such as rheumatoid arthritis, systemic lupus erythematosus and systemic sclerosis.[4] Oral symptoms are due to reduced salivary production and changes in the composition of the saliva. It is found that there is an increase in salivary lactoferrin, beta 2 microglobulin, sodium, lysozyme C and Cystatin C and a decrease in salivary amylase and carbonic anhydrase.[6] There may be presence of multiple carious lesions affecting unusual sites like anterior teeth and in the cervical region of the teeth. Patient may show unilateral parotid gland swelling and it can also become bilateral.[4]

There are many treatment options for xerostomia, namely artificial saliva, topical cyclosporine, and sialagogues (drugs or substances that increase salivary flow). Sialagogues include chewing gums, malic and ascorbic acid (very effective, but not used because they cause demineralization of tooth enamel), and the parasympathomimetic drugs. The last include Pilocarpine and Cevimeline, which bind to cholinergic (muscarinic) receptors and increase the secretion of the exocrine glands, (salivary and lacrimal glands).[7,8] Pilocarpine though is commonly used treatment option, it is contraindicated in patients with uncontrolled asthma, known hypersensitivity to Pilocarpine, patients with acute iritis and in narrow-angle glaucoma. Pilocarpine should be administered with caution in patients with SS with significant cardiovascular disease. Because Pilocarpine is

a cholinergic agonist, patients should be informed that Pilocarpine causes visual blurring and impairment of depth perception and it may have dose-related central nervous system effects along with excessive sweating.[9] As it is contraindicated in asthmatic patients and our patient had asthma, we decided to opt for the safe treatment modality which is, Artificial Saliva for the symptomatic treatment of xerostomia in our patient.

Artificial Saliva for therapeutic management of Xerostomia:

Artificial saliva functions by humidification and lubrication of the dehydrated oral mucosa. The biophysical properties of artificial saliva must be as near as feasible to natural saliva.[10] Artificial saliva contains a mixture of buffering agents, cellulose derivatives, and flavoring agents (such as sorbitol), preservatives, fluoride salts and chloride.[11] Lubrication and viscosity is provided by Carboxymethylcellulose (CMC), Viscosity/surface tension of artificial saliva is reduced using artificial mucins, Xylitol or sorbitol are added as a sweetener, To mimic the electrolyte content of natural saliva, mineral salts are added to artificial saliva. Remineralization effect is provided by fluorides also certain enzymes with antimicrobial action are added to Artificial Saliva.[11]

Artificial Saliva is used as a replacement therapy instead as treatment for dry mouth.[12] artificial saliva is used in alleviating discomfort and to help in maintaining the integrity of the teeth. The saliva substitutes must be resembling the natural saliva in terms of biophysical properties such as viscosity, mineral content, preservatives, and palatability. Artificial saliva substitute can be categorized in to two groups: Mucin and CMC based.[11] Most researchers have found the mucin-containing artificial saliva substitutes are considerably more superior to CMC containing formulations. Patients preferred a mucin-containing artificial saliva over a CMC formulation because of the better oral functioning, large retention time, and less amount required daily as found by Amal et al.[12] Mansour K. A. Assery et al in 2019 carried out a systematic review on efficacy of artificial salivary substitutes in treatment of Xerostomia. He suggested that such products reduced the symptoms of xerostomia and they should be selected as per patient's needs and concerns.[13] Another study by Dawid Lysik et al stated that Artificial Saliva supports the function of natural saliva and is useful in patients with salivary gland dysfunction and decrease in salivary secretion. It also exerts extended antimicrobial activity in patients with mucositis and dental caries.[14] In 1994 John Gibson et al also stated that other artificial saliva agents which can be for treatment of xerostomia are glycerine and lemon mouthwash where the glycerine acts as a surface-wetting agent and 2.5% citric acid as a gland stimulant. Gibson et al also added that the rheological properties of mucins and human saliva are similar but carboxymethylcellulose and polyethylene oxide solutions are non-Newtonian liquids and therefore are somewhat different. The addition of albumin to mucin solutions results in a preparation with the rheological properties which are more similar to human whole saliva.[15]

References

1. Rani S, MK Sunil, Jaskirat K, Dilip K Journal of Indian Academy of Oral Medicine and Radiology 2011;23(1):61-69
2. Stefanski AL, Tomiak C, Pleyer U, Dietrich T, Burmester GR, Dörner T: The diagnosis and treatment of Sjögren's syndrome. Dtsch Arztebl Int 2017;114: 354–61. DOI: 10.3238/arztebl.2017.0354
3. Nair JJ, Singh TP. Sjogren's syndrome: Review of the aetiology, Pathophysiology & Potential therapeutic interventions. J Clin Exp Dent. 2017;9(4):584-589.

4. Gayathry V.A, Siriwardena B.S.M.S, Jayasinghe R.D. Sjogren's Syndrome: A Case Report and Review of Literature. Sri Lanka Dental Journal 2016; 46(03: 109-115
5. Mathews SA, Kurien BT, Scofield RH. Oral manifestation of sjogren's syndrome. J Dent res 2008; Apr: 87(4):308-18.
6. Strand V Talal N Advances in the diagnosis and concept of Sjogren's syndrome (autoimmune exocrinopathy). Bull Rheum Dis. Bull Rheum Dis 1979-1980;30(9):1046-52.
7. Miranda-Rius J, Brunet-Llobet L, Lahor-Soler E, Farré M. Salivary secretory disorders, inducing drugs, and clinical management. Int J Med Sci. 2015;12(10):811-24.
8. López-Pintor RM, Fernández Castro M, Hernández G. Oral Involvement in Patients With Primary Sjögren's Syndrome. Multidisciplinary Care by Dentists and Rheumatologists. Reumatol Clin. 2015;11(6):387-394
9. M Cifuentes, P. Del Barrio-Díaz, C. Vera-Kellet. Pilocarpine and Artificial Saliva for the Treatment of Xerostomia and Xerophthalmia of Sjögren's Syndrome: A double blind control trial. British Journal of Dermatology · February 2018 Nov;179(5):1056-1061.doi: 10.1111/bjd.16442
10. Preetha A, Banerjee R. Comparison of artificial saliva substitutes. Trends Biomater. Artif Organs 2005;18(2):178–186.
11. Ekta N Ingle Artificial Saliva for Therapeutic Management of Xerostomia: A Structured Review. Journal of Oral Health and Community Dentistry 2020; 14(1) 10.5005/jp-journals-10062-0064
12. Amal A, Hussain S, Jalaluddin M. Preparation of artificial saliva formulation. ICB Pharma II 2015. 6–12.
13. Mansour K A Assery Efficacy of Artificial Salivary Substitutes in Treatment of Xerostomia: A Systematic Review J Pharm Bioallied Sci. 2019 Feb; 11(1):1–12.
14. Dawid Łysik, Katarzyna Niemirowicz-Laskowska , Robert Bucki , Gra ˙zyna Tokajuk , Joanna Mystkowska Int. J. Mol. Sci. 2019, 20, 3199; doi:10.3390/ijms20133199
15. John Gibson & Josie A. Beeley Natural and Synthetic Saliva: A Stimulating Subject, Biotechnology and Genetic Engineering Reviews 1994,12(1):39-62, DOI: 10.1080/02648725.1994.10647908

Legend Figure

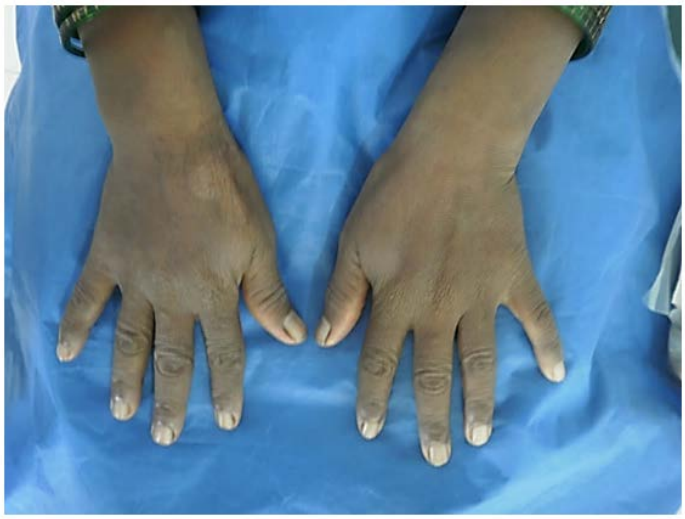

Figure 1: Swelling over small joints of hands

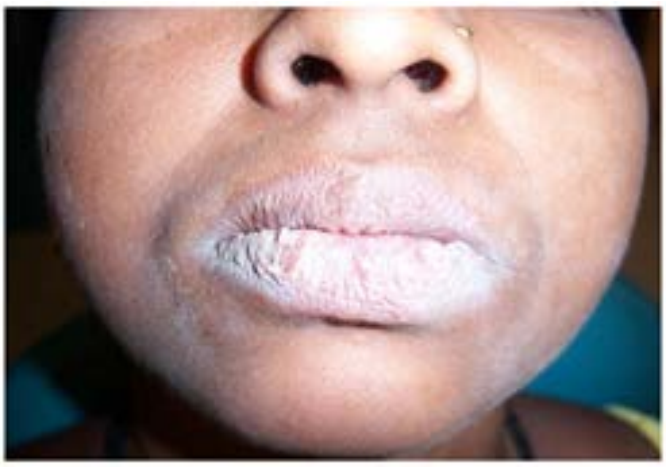

Figure 2: Dry and cracked lips

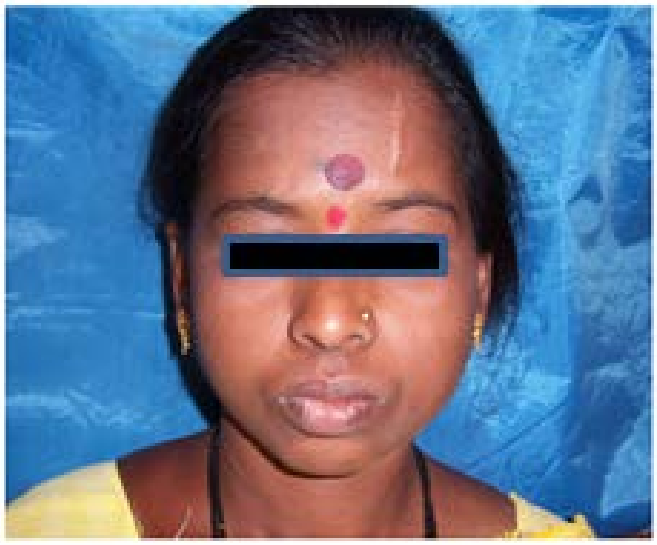

Figure 3: Bilateral swelling in the parotid region

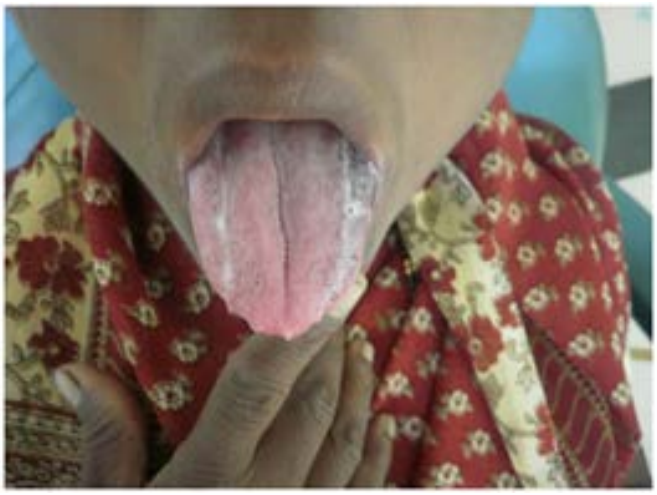

Figure 4: Thick, ropy, scanty saliva

Figure 5: Absence of salivary pool in the floor of the mouth

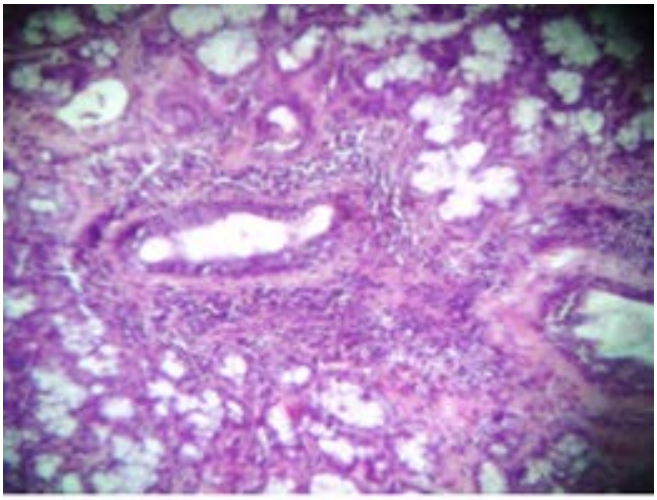

Figure 6: Histopathology of minor salivary glands