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Conservative Management of Chronic Suppurative Parotitis in Patients with Sjögren Syndrome: A Case Series

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Data Collection B
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Manuscript Preparation E
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Conflict of interest: None declared

Case series

Patients: Female, 82-year-old • Female, 77-year-old • Female, 47-year-old

Final Diagnosis: Chronic suppurative parotitis

Symptoms: Dry mouth • pus discharge • swelling

Medication: —

Clinical Procedure: Drainage

Specialty: Dentistry • Infectious Diseases

Objective: Unusual clinical course

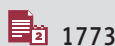
Background: Parotitis is an inflammation of the parotid gland, which can be caused by factors including infection, radiation, and hyposalivation secondary to systemic conditions, such as Sjögren syndrome, rheumatoid arthritis, or medication. Bacterial parotitis is a rare complication that can be observed in patients with hyposalivation. However, it is also observed in elderly and immunocompromised patients. Lack of continuous flushing of salivary glands and their ducts due to decreased salivary flow renders the glands prone to retrograde colonization with oral microflora. Several microorganisms have been associated with bacterial infections of the parotid glands; *Staphylococcus aureus* is the most common, accounting for 80% of cases, followed by mixed bacterial communities, including streptococci, anaerobes, and gram-negative bacilli. Bacterial parotitis presents as tenderness, swelling, and purulent sialorrhea from the salivary gland's duct. Immediate administration of broad-spectrum antibiotics, based on the results of the patient's culture and sensitivity test, has shown success in treating these cases.

Case Reports: We report 3 cases of chronic suppurative parotitis secondary to dry mouth and due to Sjögren syndrome that did not respond to oral or intravenous antibiotics and was successfully managed using conservative methods, such as the local application of superficial moist heat and periodic pus drainage by manipulating the parotid glands at dental clinics.

Conclusions: We concluded that conservative approaches, such as massaging the glands, local application of superficial moist heat, and periodic pus drainage without using antibiotics, should be considered as the first-line management of bacterial infection of the parotid glands.

Keywords: Parotitis • Salivary Glands • Sjogren's Syndrome

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/929553>



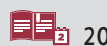
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Background

Parotitis is defined as acute or chronic inflammation of the parotid gland [1]. Factors triggering parotitis can include microbial agents (bacterial or viral), allergies, autoimmune diseases, such as Sjögren syndrome, and genetic/hereditary factors [2,3]. Acute bacterial parotitis typically presents with a sudden onset of a firm and tender swelling of the glands with occasional purulent discharge [4]. By contrast, chronic bacterial parotitis (also known as chronic recurrent parotitis [CRP]), is characterized by recurrent episodes of pain and swelling of the affected glands, with periods of remission during which the glands are without symptoms [5,6]. The exact pathogenesis of CRP is not fully understood; however, a combination of the constant reduction of salivary flow with retrograde bacterial colonization from the oral cavity may promote chronic inflammation [5,7]. Repeated episodes of parotid gland infections lead to progressive acinar destruction, fibrosis, and sialectasia [8].

Staphylococcus aureus is the most common pathogenic organism associated with bacterial sialadenitis; however, streptococcal species such as *S. pneumoniae* and *S. pyogenes* have also been implicated in bacterial sialadenitis [7,9]. Conservative interventions, including hydration, use of sialagogues, salivary gland massage, and stimulation of salivary flow with the use of sugar-free gums or sour candy, are the first steps adopted when managing bacterial parotitis [5]. The administration of systemic antibiotics may be necessary in some cases to reduce acute attacks or prevent the recurrence of CRP [5]. Surgical management as excision and drainage of the infected gland might be required in antibiotic-refractory cases or in cases with complications. In the present series, we report 3 cases of suppurative-type CRP in patients with Sjögren syndrome, in which conservative management was adopted.

Case Reports

Case 1

An 82-year-old woman with Sjögren syndrome reported non-tender bilateral swelling of the cheeks with draining discharge from both parotid glands. The patient's past medical history was significant for hypertension, asthma, congestive heart failure, hypercholesterolemia, iron deficiency anemia, gastroesophageal reflux, depression, and generalized anxiety disorder, in addition to Sjögren syndrome. The patient had a history of ovarian cancer, which was successfully managed through surgery and chemotherapy. Her medication list included enalapril, diltiazem, hydrochlorothiazide, cyclosporine ophthalmic drops, ranitidine, sertraline, cevimeline, and ferrous sulfate. Upon manipulation of the glands, a green-yellowish purulent discharge was observed bilaterally from the Stensen's duct, with symptoms being more pronounced in the right gland (Figure 1). The patient had been evaluated by the oral and maxillofacial surgery, otolaryngology, and infectious diseases departments for the recurring purulent discharge and was diagnosed with suppurative parotitis. Pus cultures were performed on 2 occasions and were shown to contain mixed bacteria, including *S. aureus*, *Klebsiella pneumoniae*, *Streptococcus viridines*, *Enterococcus*, *Pseudomonas*, and *Stenotrophomonas*. Previous treatments included courses of intravenous antibiotic therapy with ceftriaxone and ertapenem in addition to multiple trials of oral antibiotics, such as clindamycin, doxycycline, amoxicillin/clavulanate (Augmentin), trimethoprim/sulfamethoxazole (Bactrim), and ciprofloxacin, either as single agents or in combination. However, the purulent discharge never resolved, although signs and symptoms of septicemia or systemic complications had not been seen for more than 3 years. Therefore, antibiotic therapy was discontinued and

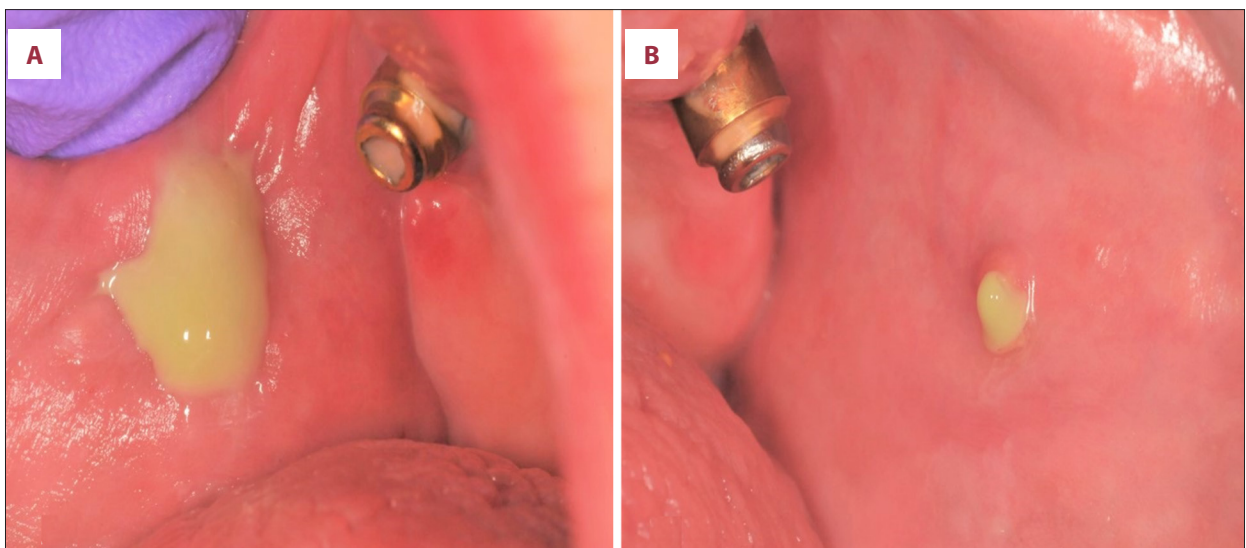


Figure 1. (Case 1) Oozing of pus from the Stensen's duct (A right and B left) after bilateral parotid gland manipulation.



Figure 2. (Case 2) A yellowish draining pus from the Stensen's duct of the left parotid gland.

followed by conservative treatment that included the nightly local application of superficial moist heat, periodic pus drainage (every 3 months) by manipulating the parotid glands at the oral medicine clinic, and use of sialagogues. The patient was followed up every 3 months for 2 years, and experienced no further episodes of CRP.

Case 2

A 77-year-old woman with Sjögren syndrome reported mild tenderness and discharge from both the parotid glands. Her medical history was significant for bronchiectasis, osteoporosis, arthritis, and Raynaud's phenomenon. Her current medications included cevimeline, nifedipine, amlodipine, cyclosporine ophthalmic drops, and multivitamins. Upon gland manipulation, a yellowish purulent discharge was observed bilaterally from the Stensen's duct, with a greater volume of discharge coming from the left side (**Figure 2**). The patient did not experience salivary gland swelling or systemic complications secondary to bacterial infection. A course of metronidazole and amoxicillin was initially prescribed, but these antibiotics failed to clear up the pus. The patient preferred to manage her symptoms with periodic manipulation of the parotid gland at the oral medicine clinic, along with the management of hyposalivation with cevimeline, moist heat, and home salivary gland massages. At the 3-month follow-up visit, the purulent discharge from the parotid glands had ceased, and a clear flow was seen from the Stensen's duct. The patient continued to use nightly local application of superficial moist heat and salivary gland massage, in addition to the use of sialagogues. She was followed up every 3 months for 2 years at the oral medicine clinic for periodic pus drainage by manipulating the parotid glands, and she experienced no further episodes of CRP.



Figure 3. (Case 3) Draining of purulent discharge from the left Stensen's duct.

Case 3

A 47-year-old woman with Sjögren syndrome reported tenderness and swelling of both parotid glands, predominantly the left gland. Her symptoms were associated with unpleasant purulent discharge from both glands. In addition to Sjögren syndrome, her medical history was remarkable for polymyalgia rheumatica and autoimmune renal failure. She was being evaluated for renal hypertrophy by a nephrologist and rheumatologist at that point. Her medications included cevimeline and prednisone. A yellowish discharge was observed bilaterally from the Stensen's duct (**Figure 3**). Signs and symptoms of systemic bacterial infection were neither reported by the patient nor detected on clinical examination. The patient was prescribed a 2-week course of amoxicillin and metronidazole. At the oral medicine clinic, the parotid glands were manipulated. The patient was advised to continue cevimeline, use moist heat, and massage the parotid glands at home. At the 2-week follow-up visit, the purulent discharge was completely resolved. Two months later, the patient reported a recurrence of purulent discharge from the left parotid gland with mild tenderness. The patient preferred conservative management of the symptoms, with periodic manipulation of the parotid gland at the clinic and management of hyposalivation with cevimeline, moist heat, and home massage. At subsequent follow-up visits, the purulent discharge from the gland cleared up with the absence of facial tenderness and systemic infection. She was followed up every 3 months for 2 years for periodic pus drainage by manipulating the parotid glands at the oral medicine clinic, while maintaining cevimeline use, moist heat application, and home massage of her salivary glands. No further episodes of CRP were reported.

Discussion

Sjögren syndrome is a chronic inflammatory autoimmune disease characterized by lymphocytic infiltration and the destruction of the exocrine glands (salivary and lacrimal glands) [10]. The classic sicca symptoms of dry eyes and dry mouth are the hallmarks of the disease [11]. In Sjögren syndrome, reduced salivary flow can promote ascending ductal infections through the colonization of the oral bacterial flora, resulting in secondary parotitis [6]. CRP can be classified into suppurative and non-suppurative types. Suppurative parotitis is caused by polymicrobial pathogens [12,13] and presents with pus discharge from the Stensen's duct upon applying pressure to the glands [2,14]. Non-suppurative parotitis is associated with numerous causes, including bacterial, viral, metabolic, genetic/hereditary, and autoimmune diseases [15]. CRP is a rare complication of Sjögren syndrome, with only a few cases reported in the literature and no established treatment guidelines [2,16]. The management of CRP mainly aims to relieve symptoms and prevent further damage to the gland's parenchyma [8,14]. The initial treatment should be conservative and may include antibiotics, adequate hydration, and good oral hygiene practices [6]. The use of antibiotics requires identification of the involved organisms by culture and sensitivity tests, followed by the administration of the proper antibiotics [6]. In addition, professional gland manipulation, gland home massage, oral sialagogues, and moist heat application are helpful adjunctive methods that may increase salivary flow and facilitate recovery [14,15]. Few reports have highlighted the outcomes of the management of secondary CRP with antibiotics in patients with Sjögren syndrome. Sugimoto et al reported the case of a 39-year-old woman with Sjögren syndrome who had several episodes of painful parotid swelling and low-grade fever. The patient was treated with prophylactic antibiotics, and responded successfully to treatment [16]. Stein et al also reported the case of a 41-year-old female patient with Sjögren syndrome who had received prophylactic antibiotics for CRP and remained in remission for 4 years [17].

In this series, and in contrast to the previously mentioned reports, we reported 3 cases of patients with Sjögren syndrome who had secondary CRP, for which conservative management approaches, with no antibiotics, successfully alleviated their signs and symptoms without the development of signs of bacteremia. The first case was given proper antibiotics according to the culture and sensitivity tests, but the infection was refractory

to antibiotics, while the other 2 patients opted not to use antibiotics. Instead, the patients were managed by receiving periodic manipulation of the parotid glands to drain the pus from the opening of the Stensen's duct at the oral medicine clinic, continuing their oral sialagogues to stimulate salivary secretion [13], and performing daily gland massaging and moist heat application at home. According to the oral medicine clinic protocol, the parotid glands are manipulated from the anterior uppermost part of the sternocleidomastoid muscles and posterior to the lower half of the earlobes. Digital pressure continues downwards below the earlobe and upwards to the tragus of the ear posterior to the masseter muscles. Patients were instructed to massage the glands at home daily after the local application of superficial moist heat. Patients were placed on a 3-month recall system for up to 2 years to ensure the continuation of the conservative management regimen, and no CRP recurrence was reported in the 3 cases. In concordance with the cases reported in this series, Patel et al described a case of CRP that persisted even after 3 courses of antibiotics. Specific pressure techniques, applied periodically, along with home care measures, facilitated recovery from the infection [15]. A literature review by Motamed et al found a 50% resolution of CRP symptoms with conservative treatment [18].

Minor surgical procedures, including sialendoscopy with intraductal steroid injection, salivary duct ligation, and tympanic neurectomy should be performed in patients who fail conservative approaches and antibiotic therapy [19]. Major surgical approaches, including superficial and total parotidectomy, should be the last resort to resolve severe, recurrent attacks of CRP that failed all the previously mentioned treatment modalities. This is mainly owing to the major complications associated with these procedures, such as facial nerve damage, Frey's syndrome, and unsatisfactory esthetic results [20].

Conclusions

Conservative management, including the periodic manipulation of the parotid glands at the dental clinic and the management of hyposalivation with either palliative measures, such as rinses for dry mouth, sugar-free gum, and the use of sialagogues (pilocarpine or cevimeline), and the home application of moist heat followed by salivary gland massage, may alleviate the signs and symptoms of some CRP cases, without the need for antibiotics.

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