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Recurrent Parotitis as a First Manifestation of Adult Primary Sjögren's Syndrome

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A 38-year-old Japanese woman was referred for evaluation of recurrent painful, bilateral parotitis. She had a 3-year history of recurrent episodes of parotitis every month, which were associated with leukocytosis and elevated levels of C-reactive protein, and which usually required antibiotic therapy. She denied having dry eyes, dry mouth, dysphagia, or arthralgia and had no other systemic symptoms. Her physical examination was unremarkable except for bilateral mildly swollen parotid glands. Laboratory tests showed a strongly positive rheumatoid factor (910 IU/ml, normal value 0-9) and anti-nuclear antibody (1:640); elevated total protein (9.2 g/dl, normal value 6.3-8.3) and immunoglobulin G (3,542 mg/dl, normal value 870-1,700) levels; normal urinalysis and complete blood counts; and a normal complement level. She was positive for anti-SS-A/Ro (134, normal value 0-9.9), but not for anti-SS-B/La (6.9, normal value 0-14.9), anti-double-stranded DNA, or anti-RNP. An ophthalmologic consultation disclosed mild corneal erosions, but showed normal Schirmer test (10 mm of wetting in 5 minutes). A chest X-ray did not show any obvious abnormalities, such as hilar lymphadenopathy or interstitial shadows. A head computed tomography scan revealed mild bilateral parotid enlargement, without sialolithiasis. The unstimulated whole salivary flow rate was 0.2 ml per 10 minutes and the stimulated flow rate was 2 ml per 10 minutes, indicating that her salivary secretion was markedly decreased in spite of the absence of oral manifestations. A labial biopsy of the minor salivary glands showed several foci of periductal lymphoid cell infiltration with glandular and ductal atrophy. Based on these findings, she was diagnosed as having primary Sjögren's syndrome [under a revised version of the

Japanese criteria (1)]. As she had had several episodes of painful parotid swelling with a low grade fever, prophylactic oral antibiotics therapy (Cefdinir at 100 mg three times per day for seven days) was started when she noticed abnormal sensation in the parotid glands. To date, she has not had any episodes of parotitis for several months.

Primary Sjögren's syndrome is a chronic inflammatory disorder characterized by lymphocytic infiltration and autoimmune destruction of the exocrine glands, with the classic symptom complex of dry eyes (keratoconjunctivitis sicca) and dry mouth (xerostomia). Salivary gland enlargement occurs in up to half of the affected patients, and the glands are usually firm, diffuse, and non-tender, with episodic swelling (2). Recurrent suppurative parotitis can be the presenting symptom in children and adolescents with this syndrome (3). However, recurrent parotitis, as in the present case, is thought to be rare as a first manifestation in adult patients with primary Sjögren's syndrome. To date, only a few reports of recurrent parotitis in adult patients with this syndrome have been reported from Japan.

The treatment of recurrent septic parotitis associated with Sjögren's syndrome has not been established. In Sjögren's syndrome, the decreased salivation or sialectasis causes a failure of duct lavage, promotes the ascent of oral bacteria to the parotid duct system, and evokes recurrent suppurative parotitis. Therefore, prophylactic antibiotic therapy might be effective for the recurrent parotitis. Indeed, Stein et al reported a 41-year-old patient with this disease who remained in remission for 4 years by receiving antibiotics treatment every night (4). The present patient complained of abnormal sensation in the parotid glands that arose before the onset of each episode; thus we started a short-term prophylactic regimen. Further clinical experience is necessary to confirm the effectiveness of this therapy.

Numerous causes for bilateral parotid swelling have been identified (5). Bilaterally infected parotid glands might be the first objective manifestation of Sjögren's syndrome, as in the present case. We recommend that Sjögren's syndrome should be considered in the differential diagnosis of recurrent parotitis even in adult patients.

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