A 46 years female presented to our OPD with severe painful swelling of both submandibular glands for 5 days. She had suffered from similar episodes of painful glandular swelling thrice in the last 6 months. The submandibular glands were swollen with prominent features of inflammation, however, the other salivary glands were not involved and the patient had no lymphadenopathy. Complete blood count revealed polymorphonuclear leucocytosis with raised ESR and the baseline biochemistry was normal. A provisional diagnosis of submandibular sialoadenitis was made and she was put on oral antibiotics. The inflammatory features regressed after 5 days of therapy though the glandular swelling persisted (Fig. 1). FNAC from the right submandibular gland at that time documented chronic non-specific submandibular sialoadenitis (Fig. 2). A detailed history revealed that the patient had some difficulties in having solid foods and she had to drink liquids to aid in swallowing dry food. She also had recurrent sensation of sand in the eyes, dry cough and a feeling of dry throat. However, she denied any definite history of decreased salivation or lacrimation. Thorough clinical examination revealed parchment like tongue with loss of filiform papillae (Fig. 3). Schirmer test using Whatman No. 41 paper suggested the presence of probable dry eyes (Schirmer I- 5mm; Schirmer II-7mm) (<5mm: confirmed dry eye; >10mm: normal; 5-10mm: probable dry eye). HIV serology, serum electrolytes and arterial blood gas analysis was normal.

Histopathological examination of the lip biopsy specimen showed heavy infiltration of the minor salivary glands with mature lymphocytes (Fig. 4). The serum specimen was positive for anti Ro/SS-A and anti La/SS-B but negative for ANF, Anti-dsDNA, RF, ANCA, Anti-Scl 70, Anti-U1RNP and AMA. Primary Sjogren’s syndrome (SS) was diagnosed as per the Revised International Classification Criteria. SS is a multisystem disorder characterised by dry eyes and dry mouth secondary to autoimmune dysfunction of exocrine glands. Parotid gland involvement is most common but submandibular and sublingual glands may also be affected. Xerotrachea and bronchial hyperreactivity may cause cough. This diversity of symptomatic expression adds to the difficulty in initial diagnosis. This case report describes a patient with SS whose sole presenting feature was bilateral and painful submandibular gland enlargement of acute onset. To the best of our knowledge there is only one previous documentation of such rare presentation of SS in world literature.1 Our idea of presenting this case is to draw the attention of physicians to the importance of considering SS in the differential diagnosis of submandibular gland enlargement.

**REFERENCE**