Soft masses occurring simultaneously in the upper and lower lips

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CLINICAL PRESENTATION

A 27-year-old man was was referred to the Department of Oral and Maxillofacial Surgery at Nagasaki University Hospital with a chief complaint of unpleasantness associated with swelling of 1.5 years' duration of the right upper and left lower lips. He was healthy and had no history of immunodeficiency or trauma involving the maxillofacial region.

Clinical examination revealed a 6 × 3 × 3 mm soft swelling in the right upper lip and an 8 × 5 × 3 mm soft swelling in the left lower lip, without cervical lymphadenopathy (Figure 1). The overlying mucous membrane was intact, and no symptoms suggestive of inflammation, such as fever, redness, or pus discharge, were observed. Routine laboratory examinations produced results within the normal range. Magnetic resonance imaging (MRI) demonstrated relatively well-circumscribed, moderately hyperintense areas on T2-weighted and gadolinium-contrast T1-weighted images of the upper and lower lips (Figure 2).

DIFFERENTIAL DIAGNOSES

Based on the clinical examinations and MRI imaging, we considered several differential diagnoses, including inflammation, mucous cyst, benign tumor, and other salivary gland diseases.

Cheilitis granulomatosa is a nonspecific granulomatous inflammatory disease that occasionally occurs. The condition is also known as Melkersson-Rosenthal syndrome or simply nonspecific inflammatory disease. Many researchers have reported that an immune response can be produced by various chronic antigenic stimuli.[1,2] The majority of patients are adults, but the phenomenon can occur at any age. The labial tissues demonstrate a diffuse, nontender, persistent swelling that may involve 1 or both lips. On rare occasions, superficial amber vesicles resembling lymphangiomas are found. When these signs are combined with facial paralysis and a fissured tongue, the clinical presentation is known as Melkersson-Rosenthal syndrome.[1,2] In addition to labial edema, swelling of other parts of the face might be seen, and cervical lymphadenopathy has been noted in rare cases. A lesion only of the lips is called cheilitis granulomatosa (of Miescher). In the present patient, a diagnosis of cheilitis granulomatosa was unlikely, because the masses in the lips were well circumscribed, in contrast to the diffuse swelling observed in cheilitis granulomatosa.

Crohn's disease (CD) is an inflammatory and immunologically mediated condition of unknown etiology that primarily affects the distal portion of the small bowel and proximal colon. This disease has oral manifestations or oral extensions, chiefly gingival hyperplasia and nodular lesions of the buccal and labial mucosa. A wide variety of disease-specific oral lesions have been described in patients with intestinal CD. These include swelling of lips, buccal mucosa swelling or cobblestoning, mucogingivitis, deep linear ulceration (usually in the buccal sulci), and mucosal tags.[3,4] Sciubba et al.[5] proposed the notion that oral manifestations of CD may be classified as an oligosymptomatic form of Melkersson-Rosenthal syndrome. Most patients with CD are teenagers when the disease first becomes evident, although another diagnostic peak of disease activity occurs in patients >60 years of age. The lesions of the gingiva appear markedly reddened and boggy and sometimes are granulomatous-appearing ulcers. The ulcers are often linear and develop in the buccal vestibule.[3,4] In the present patient, a diagnosis of CD was unlikely, because the masses in the lips were well circumscribed, in contrast to the diffuse swelling observed in CD. The reported prevalence of oral lesions in CD has varied widely. Cosnes et al.[6] found only 9 patients with oral CD among 1,805 patients with CD, giving a very low prevalence of 0.5%. However, Barnard and Walker-Smith[7] described the occurrence of oral lesions in a pediatric population with CD, and 80% of 91 patients with CD had oral findings. Even if children with aphthae are excluded from analysis, the prevalence of oral

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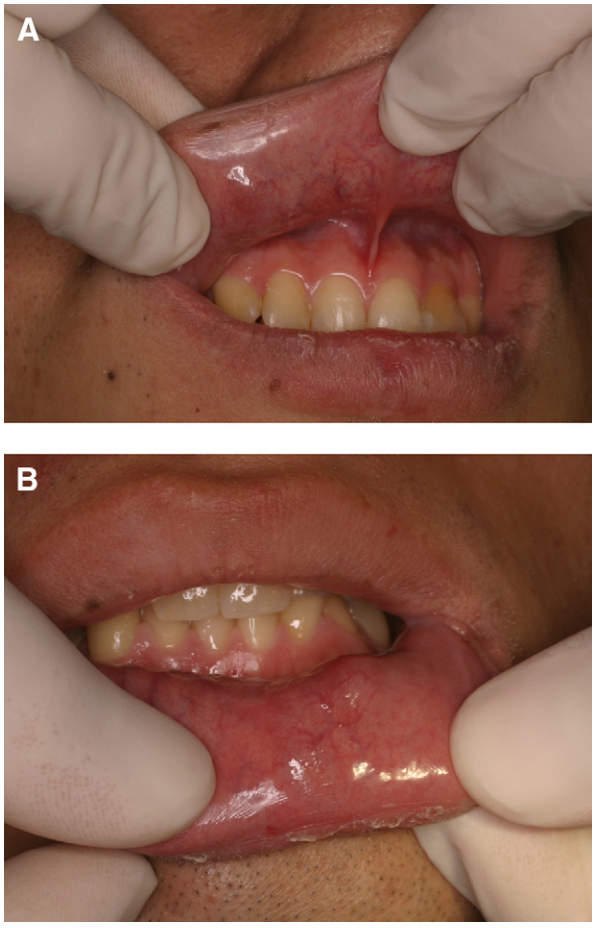

Fig. 1. Clinical examination showed relatively soft small masses in the **(A)** upper and **(B)** lower lips with no apparent symptoms suggesting inflammation.

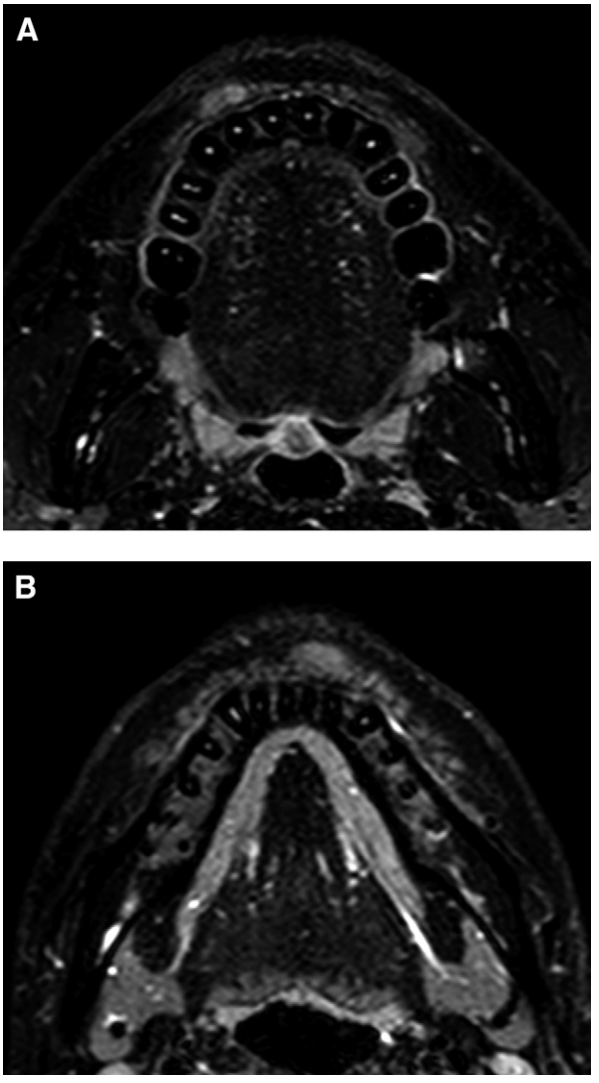

Fig. 2. Axial contrast-enhanced T1-weighted magnetic resonance images revealed well circumscribed moderately hyperintensive masses of the **(A)** upper and **(B)** lower lips.

CD lesions in that study was still high at 32%. Although the frequency with which individual types of oral CD lesions are identified has also varied between studies, Rowland et al.[8] reported that the majority of patients with oral CD had >1 finding present in the mouth.

Amyloidosis represents the extracellular deposition of fibrillar proteins (amyloid) in tissues. The primary and myeloma-associated forms of amyloidosis usually occur in older adults (average age 65 years), and a slight male predilection is present. These types of amyloidosis are caused by deposition of light chain molecules, although ~15%-20% are associated with multiple myeloma. They commonly affect the eyelid region, the retroauricular region, the neck, and the lips.[9,10] The lesions are often associated with petechiae and ecchymoses. In the present case, the lesion did not reveal any nodules, petechiae, or ecchymoses, so we considered that the lesion was not likely to be amyloidosis.

Regarding salivary gland cysts, mucocele was briefly considered as a possible differential diagnosis. This is a common lesion of the oral mucosa that results from the rupturing of a salivary gland duct and the spillage of mucin into the surrounding soft tissues. This spillage is often the result of local trauma, although there is no known history of trauma in many cases. Clinically, mucoceles typically appear as a dome-shaped mucosal swelling that can range from 1 or 2 mm to even several centimeters in size. They occur most commonly in children and young adults, but can occur in patients of all ages, including infants and older adults. The spilled mucin below the mucosal surface often imparts a bluish translucent hue to the swelling, although deeper mucoceles may be normal in color. The lesion is usually fluctuant, but some mucoceles feel firmer during palpation. The lower lip is the most common site for mucoceles, although it sometimes occurs in the upper