Journal of Dental Sciences 16 (2021) 1325–1327

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-jds.com

Correspondence

Kimura disease of the upper lip: Case report

KEYWORDS
Kimura disease;
Upper lip;
Eosinophil

Kimura disease is a rare benign reactive lymphoid proliferative disorder of unknown etiology that involves the lymph nodes and subcutaneous tissue of the head and neck regions.[1,2] Here, we reported a case of Kimura disease presented as a subcutaneous mass at the middle portion of the upper lip of a 33-year-old male patient.

This 33-year-old male patient came to our dental clinic for evaluation and treatment of a subcutaneous mass at the middle portion of the upper lip for 3–4 weeks. The mass was painless, firm, and palpable at the subcutaneous area of the upper lip. It measured approximately 0.6 cm in greatest dimension. The clinical diagnosis was either a lipoma or a minor salivary gland tumor. After discussing with the patient and obtaining the signed informed consent, the mass was totally excised under local anesthesia. The removed soft tissue specimen was sent for histopathological examination. Microscopically, it showed lymphoid hyperplasia with small- and medium-sized lymphoid follicles dispersed among the muscle bundles (Fig. 1A and B). The lymphoid follicles were composed mainly of a sheet of lymphocytes with many proliferative thin-walled capillaries and an infiltrate of eosinophils (Fig. 1C, D and E). An infiltrate of eosinophils was also discovered in the collagenous fibrous connective tissue stroma (Fig. 1F). The above-mentioned characteristic findings finally confirmed the histopathological diagnosis of Kimura disease.[1,2]

Kimura disease is a rare lymphoproliferative disease characterized by lymphoid hyperplasia with formation of lymphoid follicles and eosinophilic granulomas in the soft tissue.[1,2] Kimura disease has a male predilection with a male-to-female ratio of 14:1. It may occur at any age but the peak age is between 20 and 40 years.[1,2] Kimura disease primarily occurs in head and neck regions of Asian men and may affect major salivary glands and lymph nodes, especially in the parotid region. There are a few reports of Kimura disease cases involving the groin, limbs, eyelid, tongue, auricle, hard palate, or throat but it rarely seen in the upper lip.[1,2] The histological features of Kimura disease are very similar to those of angiolymphoid hyperplasia with eosinophilia. However, angiolymphoid hyperplasia with eosinophilia mainly affects middle-aged women in contrast to middle-aged men for Kimura disease. In addition, the patients with angiolymphoid hyperplasia with eosinophilia rarely have peripheral blood eosinophilia, increased serum IgE, and lymphadenopathy, but the patients with Kimura's disease are typically associated with the aforementioned three characteristic clinicopathological findings.[1,2] Peripheral blood eosinophilia, increased serum IgE, and lymphadenopathy are very helpful for diagnosis of Kimura disease. However, the final diagnosis of Kimura disease relies on a pathological examination.[1,2] Although immunohistochemical stains are frequently used for histopathological diagnosis of tumors or lesions at the head and neck region and identification of tumor cell origin,[3–5] they are not necessary for the diagnosis of Kimura disease. Surgery is the usual primary approach for diagnosis and therapy for Kimura disease.

https://doi.org/10.1016/j.jds.2021.02.004

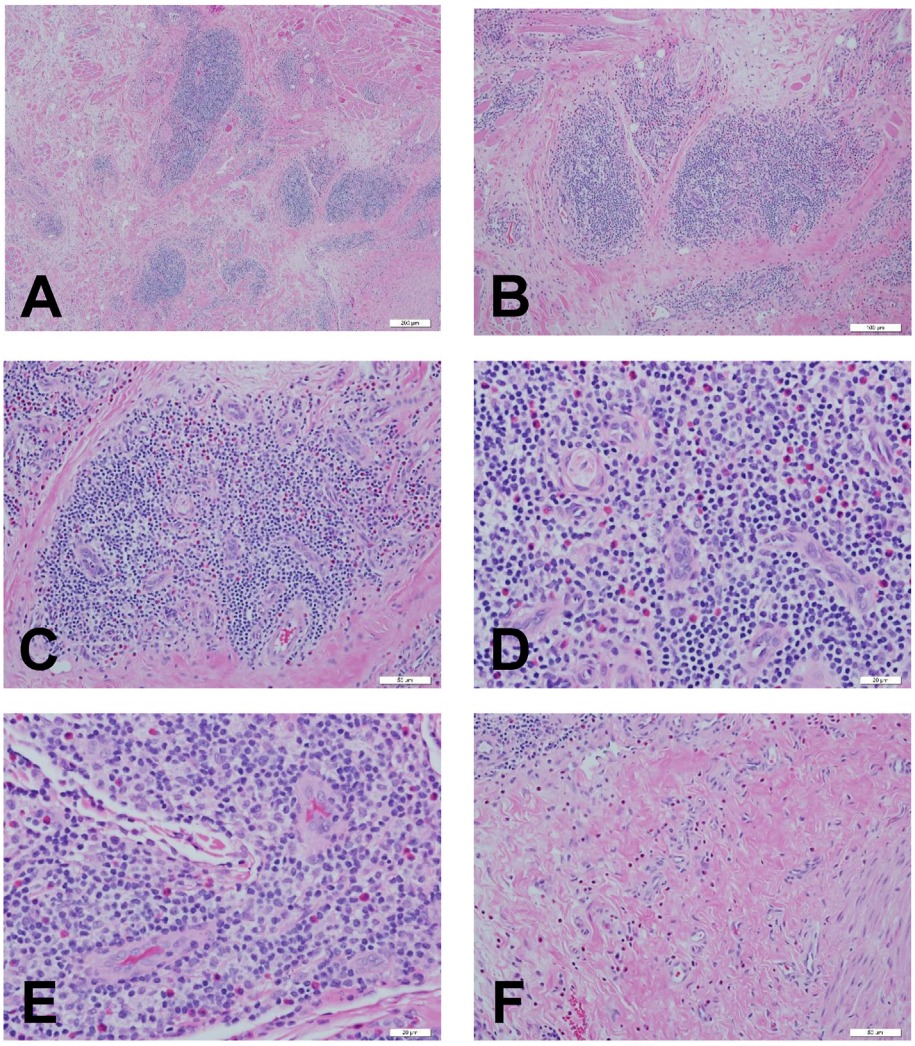

Figure 1 Histopathological microphotographs of our case of Kimura disease. (A and B) Low-power microphotographs showing lymphoid hyperplasia with several small- and medium-sized lymphoid follicles dispersed among the muscle bundles (Hematoxylin and eosin stain or H&E; original magnification; A, 4×; B, 10×). (C, D and E) Medium- and high-power microphotographs demonstrating several lymphoid follicles composed mainly of a sheet of lymphocytes with many proliferative thin-walled capillaries and an infiltrate of eosinophils (H&E; original magnification; C, 20×; D, 40×; E, 40×). (F) Medium-power microphotograph exhibiting an infiltrate of eosinophils in the collagenous fibrous connective tissue stroma (H&E; original magnification; 20×).

Other treatment modalities for Kimura disease include regional or systemic steroid therapy, cytotoxic therapy, and radiation. The Kimura disease has an excellent prognosis, although it may recur locally.[1,2]

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

References

1. Zhang X, Jiao Y. The clinicopathological characteristics of Kimura disease in Chinese patients. *Clin Rheumatol* 2019;38:3661—7.
2. Zhang L, Yao L, Zhou WW, Ma JN, Zhang CQ. Computerized tomography features and clinicopathological analysis of Kimura disease in head and neck. *Exp Ther Med* 2018;16:2087—93.
3. Hsu CW, Tseng CH, Wang WC, Chen YK. Alveolar soft part sarcoma of tongue in a 3-year-old Taiwanese. *J Dent Sci* 2019;14:325—7.

4. Chen CY, WangWC, Tseng CH, Su CW, Chen YK. Clinicopathological study of 13 cases of intraoral soft tissue metastatic carcinomas. *J Dent Sci* 2020;15:92—5.
5. Hwang MJ, Huang BW, Lee YP, Chiang CP. Langerhans cell histiocytosis in an old man — case report. *J Dent Sci* 2021;16:558—60.

Ming-Jay Hwang[†]
Department of Dentistry, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan
Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan

Julia Yu-Fong Chang[†]
Andy Sun
Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan
Graduate Institute of Oral Biology, School of Dentistry, National Taiwan University, Taipei, Taiwan

Chun-Pin Chiang*
Department of Dentistry, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan
Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan
Graduate Institute of Oral Biology, School of Dentistry, National Taiwan University, Taipei, Taiwan

*Corresponding author. Department of Dentistry, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, No. 707, Section 3, Chung-Yang Road, Hualien 970, Taiwan.
E-mail address: cpchiang@ntu.edu.tw (C.-P. Chiang)

Received 16 February 2021
Available online 4 March 2021

[†] These two authors have equal contribution to this study.