Recurrent Kimura disease of the parotid gland

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ABSTRACT

Kimura disease is a rare chronic inflammatory disorder affecting young males of eastern countries. There are about 200 cases reported in the English literature, with <60 cases involving the parotid gland. It is characterized by abnormal proliferation of lymphoid follicles with eosinophil-rich infiltrate and peripheral eosinophilia. The disease presents as painless cervical lymphadenopathy or subcutaneous masses in the head and neck region. Although recurrence is common, radical surgery is not recommended, as the disease is essentially benign.

Key words: Eosinophilia, Kimura disease, parotid

A 35-year-old Indian male presented with a 10 cm \times 10 cm painless swelling in the left parotid region for 2 years. He had undergone wide local excision (WLE) of the swelling in the same area 2 years back, and the histopathological examination (HPE) suggested Kimura disease (KD). The swelling had recurred 14 months following WLE. Routine hemogram showed eosinophilic leukocytosis (total leukocyte count, TLC: 14,000/mm³, with 42.8% eosinophils). Computerized tomography scan of the face showed a left-sided soft tissue swelling of the cheek involving the parotid gland and adjoining subcutaneous fat plane. Fine-needle aspiration cytology showed eosinophilic infiltrate. Nerve sparing superficial parotidectomy was done [Figure 1]. HPE revealed follicular hyperplasia and eosinophilic infiltration of lymph nodes suggestive of KD [Figure 2]. One month post-operatively TLC and the differential eosinophil count had normalized. Follow-up at 2nd and 6th months post-operatively revealed the patient to be asymptomatic and clinically free of any disease.

Kimura disease was first reported in 1937 by Kimm and Szeto [1] from China, and later in 1948 by Kimura et. al.[2] The etiology is unknown; allergy, infection, or an autoimmune process may have a role to play. The basic pathology is abnormal proliferation of lymphoid follicles, with eosinophil-rich infiltrate and peripheral eosinophilia. In the reported case, eosinophilia resolved after surgical excision of the tumor. KD is most commonly seen in young males of eastern countries in the third decade of life, which is consistent with the demographic profile of our patient [3,4]. The salivary glands, skin and lymph nodes in the head and neck region are commonly involved, especially the periauricular, parotid and submandibular regions. In our case, only the parotid gland was

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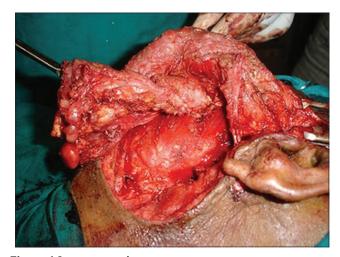


Figure 1 Intra-operative appearance

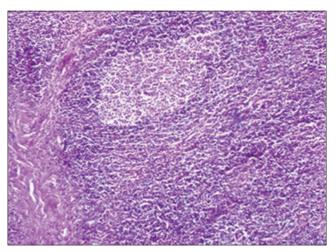


Figure 2 Microscopic appearance: eosinophilic infiltration of lymph nodes (H and E, ×100)

involved. There is an unexplained association with nephrotic syndrome in up to 20% cases [5]. Fortunately this was not seen in our patient. Surgical excision is the treatment of choice for KD. Recurrence is common, and re-do surgery, or systemic therapy may be required. Steroids are recommended

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CLINICAL IMAGE

in KD associated with nephrotic syndrome. Cyclosporine induces remission, but recurrence is observed once therapy is stopped. Radiation may be considered in recurrent cases with disfiguring lesions [6]. Although our patient had a recurrence, we did not give any adjuvant therapy, which we kept reserved in case a second recurrence occurred.

Kimura disease should be considered as a differential diagnosis in Oriental patients presenting with parotid swelling and eosinophilia. Although recurrence and lymphadenopathy may suggest a malignant etiology, KD is essentially a benign disease and nerve sparing parotidectomy is adequate surgery for KD affecting the parotid gland.

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Author's Contributions

SP, SS and AD contributed to the literature search whereas SS, DM and AD helped in drafting the manuscript. All authors have read and approved the final version of the manucript.

Consent

The authors certify that a written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

Competing Interest

Nil

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