Intraparotid Kimura's disease: An unusual case presentation

Sir,

Kimura's disease (KD) is a chronic inflammatory disorder of obscure etiology that most commonly presents as painless lymphadenopathy or subcutaneous masses in the head and neck region [1]. Histologically, KD is characterized by eosinophilic lymphoid granulomas, eosinophilic abscesses with vascular proliferation and variable degrees of fibrosis. For years, KD was believed to be identical to or part of the same disease spectrum as angiolymphoid hyperplasia with eosinophilia (ALHE). Recent reports, however, have confirmed that the two are, in fact, separate entities [2]. Parotid gland involvement is not very common but, nevertheless, may occur as in the present case.

We present a case of 67-year-old male presented with swelling on the left side of the face for 1 year. The swelling did not increase in size and was painless. There was no history of fever or weight loss. He did not have associated cough, hemoptysis, upper respiratory tract, or urinary complaints. On examination, multiple, irregular, and tan brown soft tissues measuring 6 cm × 3 cm × 1 cm were shown. All the vitals were within normal range. Histopathological examination (HPE) showed extensive areas of fibrosis with a scant remnant of parotid tissue (Fig. 1a). Lymphoid follicles with a prominent germinal center and eosinophilic microabscesses were shown (Fig. 1b and c). In addition, small vessels with prominent endothelial lining were also noted (Fig. d). HPE diagnosis of intraparotid KD was signed out. Fine-needle aspiration cytology was suggestive of chronic parotitis. The pre-operative investigations showed peripheral blood eosinophilia with absolute eosinophil count of 1020/mm^3. After which, superficial parotidectomy was performed, which was uneventful.

KD was established as a separate entity in 1979 by Rosai *et al*. [3]. It is commonly seen in young Asian males with the predilection of the head and neck region [2]. KD commonly presents as lymphadenopathy and subcutaneous swelling. However, parotid gland involvement is rare, simultaneous involvement of lymph nodes and parotid gland has also been reported [2]. Previously, KD was considered as a variant of ALHE, however, now both are known as distinct entities with different etiopathogenesis. The differences between KD and ALHE are highlighted in Table 1.

KD is a reactive immune disorder characterized by the presence of peripheral eosinophils, increased mast cells, and increased levels of interleukin-5 and Immunoglobulin E (IgE), which imply an abnormal T-cell stimulation akin to a hypersensitivity-type reaction [1]. Histopathologically, the constant features reported are preserved nodal architecture, florid germinal center hyperplasia, eosinophilic infiltration, and postcapillary venule proliferation. Frequent features comprise of sclerosis, polykaryocytes, vascularization of the germinal centers, proteinaceous deposits in the germinal centers, necrosis of the germinal centers, and eosinophilic abscesses. The solitary

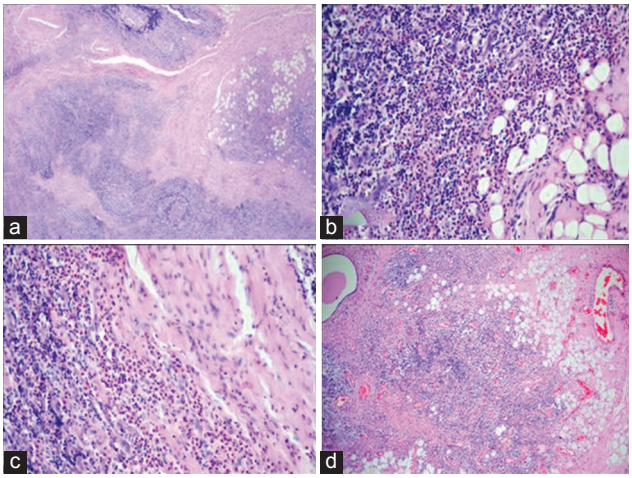

Figure 1: (a) Histopathology shows extensive areas of fibrosis with scant remnant of parotid tissue (H and E, ×10). (b and c) Lymphoid follicles with prominent germinal center and eosinophilic microabscesses (H and E, ×400). (d) Small vessels with prominent endothelial lining (H and E, ×10)

Table 1: Differences between Kimura and ALHE [2,6]

Parameter	KD	ALHE
Pathogenesis	Allergic inflammatory process	Benign vascular proliferative disorder
Sex distribution	Common in young males	Females in the 3rd–4th decade
Racial distribution	Common in Asian males	No racial predilection
Site	Common in head and neck	Generalized distribution
Clinical presentation	Regional lymphadenopathy, localized subcutaneous mass, peripheral eosinophilia, elevated serum IgE	Erythematous papules or nodules with tenderness and pruritis
Histopathology	Lymphoid follicles with prominent germinal centers, eosinophilic abscesses, fibrosis, and flattened blood vessel endothelium	Florid proliferation of blood vessels and mild-to-moderate lymphoid hyperplasia, variable eosinophilic infiltration, eosinophilic microabscess absent, plump/histiocytoid blood vessel endothelium

ALHE: Angiolymphoid hyperplasia with eosinophilia, KD: Kimura's disease, IgE: Immunoglobulin E

rare feature is the progressive transformation of the germinal centers. Immunochemistry reveals the presence of IgE reticular network in germinal centers and IgE-coated non-degranulated mast cells [4].

The histopathological picture of lymphoid proliferation and eosinophilia gave rise to a differential diagnosis of Hodgkin's lymphoma, angioimmunoblastic T-cell lymphoma, Langerhans cell histiocytosis, and parasitic lymphadenitis. However, the absence of classic Reed–Sternberg cells and/or its variants, atypical lymphocytes, Langerhans cells, and parasitic remnants aided in separating our case from the aforementioned entities [5].

Surgical excision is the first line of treatment for symptomatic cases. Topical and systemic corticosteroids have also been effective, and in patients resistant to steroids, radiation therapy has been used [2].

Thus, to conclude, we can say that KD is no longer considered a variant of ALHE. KD, though rarely encountered, must be kept in mind in the differential diagnosis of a parotid mass.

Shailja Puri

From Department of Pathology, SRL laboratory, Shimla, Himachal Pradesh, India

Correspondence to: Shailja Puri, Department of Pathology, Consultant Pathologist and Lab Head, SRL Ltd., Shimla Himachal Pradesh, India.
E-mail: drshailjadoe_11@ymail.com

Received - 15 April 2018
Initial Review - 18 May 2018
Accepted - 01 August 2018

REFERENCES

1. Meningaud JP, Pitak-Arnnop P, Fouret P, Bertrand JC. Kimura's disease of the parotid region: Report of 2 cases and review of the literature. J Oral Maxillofac Surg 2007;65:134-40.
2. Sah P, Kamath A, Aramanadka C, Radhakrishnan R. Kimura's disease - An unusual presentation involving subcutaneous tissue, parotid gland and lymph node. J Oral Maxillofac Pathol 2013;17:455-9.
3. Rosai J, Gold J, Landy R. The histiocytoid hemangiomas. A unifying concept embracing several previously described entities of skin, soft tissue, large vessels, bone, and heart. Hum Pathol 1979;10:707-30.
4. Hui PK, Chan JK, Ng CS, Kung IT, Gwi E. Lymphadenopathy of Kimura's disease. Am J Surg Pathol 1989;13:177-86.
5. Chen H, Thompson LD, Aguilera NS, Abbondanzo SL. Kimura disease: A clinicopathologic study of 21 cases. Am J Surg Pathol 2004;28:505-13.
6. Ramchandani PL, Sabesan T, Hussein K. Angiolymphoid hyperplasia with eosinophilia masquerading as kimura disease. Br J Oral Maxillofac Surg 2005;43:249-52.

Funding: None; Conflict of Interest: None Stated.

How to cite this article: Puri S. Intraparotid Kimura's disease: An unusual case presentation. Indian J Case Reports. 2018;4(4):338-339.

Doi: 10.32677/IJCR.2018.v04.i04.029