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Kimura's Disease of the Parotid Region: Report of 2 Cases and Review of the Literature

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Kimura's disease (KD) is an uncommon chronic inflammatory disorder of unknown etiology. Clinically, it presents as solitary or multiple subcutaneous nodules, predominantly in the head and neck region, typically in the preauricular region, forehead, and scalp.¹⁶ Other localizations such as lacrimo-orbital involvement and upper extremities have been reported.^{1,6} The soft tissue localization is often associated with regional lymphadenopathy and, occasionally, with enlargement of the major salivary gland.²⁻⁵ Sporadic cases in non-Asian peoples with KD have been previously reported,^{4,6,7} but there are no reports of patients from islands in the Southwest Indian Ocean. We report 2 cases of KD with hypereosinophilia in patients who are natives of Madagascar and Mauritius. Our report shows that KD should be considered even in non-Asian patients in the diagnosis of hypereosinophilia-associated disease with head and neck subcutaneous mass.

Report of Cases

CASE 1

A 29-year-old man who was a native of Madagascar with no past medical history gradually developed a large, firm mass over the right preauricular region involving the right

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zygomatic and temporal areas over 3 months (Fig 1). The cervical lymph nodes were not palpable; physical examination was otherwise unremarkable. Laboratory results were a marked peripheral hypereosinophilia. Because there was no evidence of allergy or parasitic infestation and because the patient had not taken prior medications, hypereosinophilic syndrome was first considered due to the markedly elevated blood eosinophil count. However, the absence of internal organ involvement made this diagnosis questionable. Computed tomography (CT) scan revealed an expansively illdefined subcutaneous mass occupying the parotidomasseteric region: there was no bone involvement. Nevertheless. it was not determined whether the lesion originated from the parotid gland (Fig 2). Magnetic resonance imaging (MRI) showed that the lesion infiltrating the subcutaneous fatty tissue had inhomogeneous low signal intensity on both T1- and T2-weighted images; the lesion margins were illdefined without a clear plane of cleavage between the mass and the adjacent parotid gland that was displaced and compressed. No evidence of the invasion of masseter muscle and underlying bone could be seen (Fig 3). Under general anesthesia, surgical removal of the mass with preservation of the facial nerve was performed via rhytidectomy. Pathologic study of the specimen showed a marked reactive follicular hyperplasia with prominent follicles that were surrounded by fibrous tissue. Some follicles contained eosinophilic proteinous material; a few demonstrated folliculolysis. The interfollicular infiltrate was rich in plasma cells and eosinophils that formed scattered eosinophilic microabscesses associated with granulomatous inflammation. Thin-walled vessels were numerous and often grouped in small foci; their endothelial lining was neither epithelioid nor vacuolated. Reed-Sternberg cells, atypical lymphocytes, or Langerhans cells were not identified. Special stains including periodic acid-Schiff, Giemsa, and Brown and Brenn failed to show any infectious agents (Figs 4-6). Therefore, the definite diagnosis was KD. The patient's condition has remained satisfactory throughout the 1-year follow-up period.

CASE 2

A 25-year-old Mauritius native male presented with a soft, mobile, painless, and slowly growing, approximately for 18 months, mass over the right parotid region in 1991. With the exception of hypereosinophilia, there was no significant medical history. Lymph nodes from the left spinal, left suprahyoid, left suboccipital, and left axillary groups, as well as from the right inguinal group, were enlarged. CT scan showed a well-circumscribed 8.5×3.5 cm diameter



FIGURE 1. Photograph of the patient case 1 who presented with a 3-month history of progressive swelling of the right preauricular regions involving the right zygomatic and temporal areas.

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mass that originated from the right parotid gland. The patient benefited from right superficial parotidectomy at another hospital. Histologic examination demonstrated lymphoid hyperplasia with vascular proliferation and eosinophilic infiltration; therefore, KD was diagnosed. Postoperative facial paralysis was encountered which was an unsatisfactory surgical outcome. In 1999, the patient was referred to us for consultation because of recurrence of KD as a right preauricular lymphadenopathy confirmed by CT scan (Fig 7). To avoid further injury to the facial nerve, the mass was excised via an intraoral approach. The diagnosis of KD was confirmed pathologically. The postoperative course was uneventful. However, in early 2004 the patient presented with a 4 cm diameter mass in the left retromandibular area. CT scan and MRI revealed hyperplasia of the inferior pole of the left parotid gland consistent with lymphoid hyperplasia (Figs 8, 9). A left superficial parotidectomy via a retromandibular approach was performed. Histologically, the mass contained the prominent lymphoid hyperplasia compartmentalized by fibrous connective tissue; the interfollicular infiltrate was rich in plasma cells and eosinophils that formed eosinophilic microabscesses. The final diagnosis was KD. At the time of last follow-up there had been no recurrence for 1 year. There was no renal complication.

Discussion

KD is an uncommon chronic inflammatory disorder involving subcutaneous tissue, predominantly in the head and neck region. This disease is most frequently associated with regional lymphadenopathy and/or salivary gland involvement.^{3-5,8-10} Although KD has long been described as a distinct entity since the original report by Kimura,²⁻⁵ it is often confused with angiolymphoid hyperplasia with eosinophilia (ALHE) in many reports.¹¹⁻¹⁴ A long list of synonyms for KD has added to the confusion. However, KD and ALHE are quite different and should be distinguished using clinical and pathologic features. A brief summary of distinctive features between these entities is presented in Table 1. KD has a predilection for young to middleaged Asian men; however, some cases have been reported in non-Asian patients. Although both our patients are natives of islands located in the Southwest Indian Ocean, many people from these countries are partially of Asian descent. It is generally assumed that there is a genetic predisposition for KD.

Current theories on precise causation have been the subject of debate. Because of the correlations of KD with immunities, it seems suitable to classify this disease in a category of the reactive immune disorders in etiology, and not with tumors.⁸⁻¹⁶ Various leukocytes have been advocated in the disease etiopathogenesis, including mast cells,^{17,18} eosinophils,¹⁹ and macrophages.^{20,21} Recent studies have also suggested that the expression of activated CD4(+) Thelper 2 (Th2) cells,^{10,22} as well as the overexpression of their cytokines, such as granulocyte-



FIGURE 2. A and B, Axial CT scan of case 1 showing an expansive, ill-defined, homogeneously hypoattenuated mass occupying the right parotidomasseteric region and extending posteriorly toward the posterior border of mandilar ramus. No bony involvement was evident.

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FIGURE 3. MRI of case 1 showing ill-defined inhomogeneous hypointense lesions located in the subcutaneous tissue of parotidomasseteric region without a clear plane of cleavage between the mass and the adjacent parotid gland appearing displaced and compressed. No evidence of the invasion of masseter muscle and underlying bone can be seen.

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macrophage colony-stimulating factor, tumor necrosis factor- α , interleukin (IL)-4, IL-5, eotaxin, and RANTES,^{23,24} may play an important role in the disease development by regulating Ig E production and eosinophilic recruitment. Moreover, a clonal T-cell population may contribute to disease development and recurrence.²⁵ Some antigens responsible for the reactive induction of the immune system have been proposed; such as *Candida albicans*,¹⁶ Epstein-Barr virus,²⁶ and human herpes virus-8.²⁷



FIGURE 5. Photomicrograph of case 1 showing germinal centers with lack of distinctive large epithelioid endothelial cells and vascular proliferation. The interfollicular infiltration was rich in plasma cells and eosinophils forming eosinophilic microabscesses. (Hematoxylin-eosin stain; magnification ×200.)

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Clinically, KD lesions simulate neoplasms in the head and neck region,^{2-5,10,14,16} sometimes in the upper limbs.^{28,29} The lesions present as deeper masses rather than isolated or grouped red papules, plaques, or mucocutaneous lesions that are commonly seen in ALHE.¹⁵ Both diseases can cause proptosis, lid swelling, ocular dysmotility, or a palpable mass when encountering the orbit and ocular adnexa. Nevertheless, KD has been described in the orbit, eyelids, and lacrimal gland more frequently than ALHE.¹¹ For the



FIGURE 4. Photomicrograph of case 1 showing lymph node-like tissues with markedly reactive follicular hyperplasia and granulomatous inflammation interspersed with normal fatty tissue. (Hematoxylin-eosin stain; magnification ×25.)

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FIGURE 6. Photomicrograph of case 1 showing numerous and prominent thin-walled vessels with lack of epithelioid or vacuolated endothelial cells. There was no atypia in the lymphoid cell population. (Hematoxylin-eosin stain; magnification ×400.)

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FIGURE 7. CT scan of case 2 showing a recurrent mass as a right preauricular lymphadenopathy. The patient, subsequently, underwent the excision via an intraoral approach as the second operation.

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non-Asian with KD, some authors have reported its localization in preauricular subcutaneous tissue and cervical lymph nodes of male Caucasians; most had peripheral hypereosinophilia.⁴ Lymph nodes and major salivary glands, especially parotid glands, are commonly affected, resembling malignancies.^{2,3} Interestingly, no bilateral parotid masses have been previously documented, as has been shown in our second case. Swelling of the extraocular muscle,³⁰ juvenile temporal arteritis,³¹ and associated coronary artery disease³² are also grouped in accessory lesions of KD, including sleep apnea in cases of laryngeal involvement.³³

Many patients with KD also develop allergic conditions and renal involvement. Asthma, chronic urticaria/ eczema, pruritis, rhinitis, cutaneous eosinophilic vascu-



FIGURE 8. CT scan of case 2 before the third operation shows a lesion in the region of the left parotid.

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FIGURE 9. MRI of case 2 before the third operation revealed hyperplasia of the inferior pole of the left parotid gland consistent with ymphoid hyperplasia.

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litis, and alopecia areata are frequently associated.^{2,5,14} The coexistences of KD and other cutaneous lesions have also been reported, such as lichen amyloidosus. It is believed that the inflammatory process of KD results in the basal layer damage necessary for collision occurrence of the later lesions.³⁴ Also, it has a high frequency of an association with nephrotic syndrome; a handful of KD pediatric patients with renal diseases or who require dialysis have been reported.^{10,22,35,36} This renal involvement has been attributed to the deleterious effects of eosinophil granules and possibly to microemboli from the heart in patients presenting with fibroplastic endocarditis

Table 1. THE COMPARISON BETWEEN KD AND ALHE

	Kimura's	
	Disease	ALHE
Gender	Predominantly	Predominantly
	male	female
Ethnic group	Predominantly Asian	Asian and non-Asian
Localization	Deep, subcutaneous	Superficial, dermal
Clinical presentation	Nodule	Papule
Lymphadenopathy	Frequent	Rare
Salivary gland involvement	Frequent	Rare
Hypereosinophilia	Frequent	Rare
Serum Ig E	Elevated	Normal
Predominant cellular component	Lymphoid infiltrate	Vascular hyperplasia
Feature of endothelial cells	Flat	Epithelioid and/or vacuolized

Meningaud et al. Kimura's Disease of the Parotid Region. J Oral Maxillofac Surg 2007. or eosinophilic myocarditis. Most secondary forms are usually caused by an immunoallergic process leading to the deposit of immune complexes in glomeruli. The effects of polynuclear eosinophils could also be caused by the release of cytokines and other mediators. Additionally, inflammatory process in KD may be involved in refractory hypertension and anemia in patients with end-stage renal disease requiring dialysis,⁹ steroid-resistant nephrotic syndrome,³⁶ as well as renal insufficiency.³⁷ Fortunately, both our patients had no history of or were not associated with renal disease. Nevertheless, in our opinion, KD patients should be referred for other physical evaluation, particularly for nephropathy.

Imaging evaluation of soft-tissue lesions has undergone a dramatic evolution with the advent of CT and MRI imaging, which are very useful for definite preoperative diagnosis. Ultrasonogram (US) can also be useful. CT scan provides the information of this soft tissue lesion without any bony involvement. On CT scans with contrast enhancement this lesion appears as a homogeneous, slightly hyperattenuated mass. MRI will differentiate its precise nature from other soft tissues tumors. This lesion tends to be heterogeneous-hypointense, sometimes slightly hyperintense on T1- and T2-weighted imagings. Additionally, it will show heterogeneous enhancement after administration of contrast media. Its discrepancies of enhancement degree, by nature, may be attributable to differing degrees of fibrosis and vascular proliferation.³⁸ The lesions always present a subcutaneous woollyechotexture mass with well- or ill-defined border on US.³⁹ In addition, on gray-scale US, hypoechoic and round features with normal hilar architecture and homogeneous internal echoes are always revealed in nodal lesions. The prominent intranodal vessels with a hilar pattern and low intranodal resistance are always revealed on Doppler US, as well as soft tissue; parotid lesions also show low-resistance vascularity within.⁴⁰

Concerning the presentation in our first case, the exact plane of the lesions should be established to differentiate them from tumors in other origins. The differential diagnosis in emphasis was made in groups of soft tissue tumors, such as lipoma, rhabdomyoma, neurilemmoma, or schwannoma, and lymphoproliferative or granulomatous lesions, including Mikulicz's disease (sialolymphadenitis) or Castelman's disease.^{16,41} Hyperplasia and benign tumors of subcutaneous vascular proliferations should also be in the differential list, including ALHE, bacillary angiomatosis, intravascular papillary endothelial hyperplasia, Kaposi's sarcoma, pseudo-Kaposi's sarcoma, and various types of hemangiomas. As demonstrated in the second case, when lymph nodes are involved in the submental, submandibular, and upper cervical regions, as well as within the parotid gland, it mimics malignancy. The differential diagnosis with reactive lymphadenopathy, subcutaneous soft tissue tumors with lymph node involvement, Hodgkin's lymphoma, dermopathic lymphoma, parotid tumor with nodal metastasis, and Mikulicz's disease or Castelman's disease should not be excluded.^{40,41} Differentiation of Langerhans cell histiocytosis and salivary gland neoplasms from KD is also crucial in the region of the parotid gland.⁴²

Diagnosis is achieved by excisional biopsy, which is also the therapy of choice. Microscopically, as compared with ALHE, KD shows a marked lymphoproliferation with prominent germinal center and some folliculolysis. Also, KD lacks distinctive large epithelioid endothelial cells. Instead, the angioproliferation involved postcapillary venules presenting with thinwalled vessels^{4,15} are often accompanied by a characteristic inflammatory infiltration, forming eosinophilic microabscesses. In nodal specimens, florid germinal centers, vascularized germinal centers, increased paracortical post-capillary venules, eosinophilic infiltration, and fibrosis are commonly found. Warthin-Finkeldey polykaryocytes are often positive for lymphoid and macrophagic markers. Fine needle aspiration cytology is characterized by an admixture of significant numbers of eosinophils, fragments of collagenous tissue, epithelioid endothelial cells, and occasional polykaryocytes (Warthin-Finkeldey-type giant cells) against a background of a polymorphous lymphoid population. Therefore, cytology can be a useful adjunct diagnostic tool in KD.43,44

Treatment options range from conservative observation for asymptomatic patients to surgical excision, steroid therapy, and radiotherapy for the symptomatic patient. The lesion is benign but may be persistent and difficult to eradicate. Multiple treatment modalities have been proposed for KD, each with limited success or undesirable side effects and recurrence is common. Treatments include oral corticosteroids,^{7,32,41} oral retinoids,⁴⁵ oral pentoxifylline,⁴⁶ cyclosporine,^{34,47,48} surgical excision,³ radiotherapy,^{26,41,49} and laser therapy.³³ Overall prognosis is favorable, but multiple relapses are possible. There is no evidence of malignant transformation and occasionally spontaneous resolution occurs.⁷

Similar to our opinion, some reports have suggested that complete surgical excision with/without initial high-dose corticosteroid whenever feasible is the preferred treatment for a well-circumscribed lesion, despite a high recurrence rate.^{3,50} With great emphasis on diffusely infiltrative lesions, complete extirpation is virtually impossible and may result in recurrence.¹⁹ When surgery is not possible or in situations of recurrence, conservative treatment with either corticosteroids^{7,14,32,41} or radiation^{26,41} often produces a favorable response. On the other hand, many internists advocated medical management (eg, corticosteroids).^{7,14,32,41} Some immunomodulators, such as pranlukast or suplatast tosilate, have been observed to support steroid tapering by the possible immunomodulatory mechanism of IL-4 production of Th2 cells.^{51,52} Because of their effect on the Th2 cells, cyclosporine,47,48 interferon-alpha,53 and oral pentoxifylline⁴⁶ may also be introduced in cases of steroid-resistant KD subcutaneous masses or ulcers. If the degree of improvement is unsatisfactory or drug resistance occurs, the more radical excision should be considered later. Nicotine, which is an effective treatment of other neutrophil-associated skin disorders (such as pyoderma gangrenosum and orogenital ulcerations caused by Behcet's disease) may be attributable to prominent eosinophilic infiltration such as KD skin lesions.⁵⁴ Prospective, randomized clinical trials, however, are needed to answer whether these medicaments are a substitute for, or adjunct to, surgery.

Renal involvement of KD subsides most of the time after administration of corticosteroid.⁹ In steroid-resistant cases, KD masses must be sacrificed.³⁶ Care should be taken in KD cases of renal transplantation because both chronic graft rejection and KD involve the Th2-dominant immune response, which may lead to secondary renal graft failure.¹⁰

KD is an uncommon chronic inflammatory disease presenting as a subcutaneous swelling typically in the head and neck area. It has been described more often in Asians. However, it does occur in non-Asians with a similar clinicopathologic presentation. Our 2 cases suggest that KD is no longer just a disease from Oriental countries, although most cases have been from there. KD is a distinctive entity with elusive etiology. It has characteristic histologic features that are important to recognize, including follicular hyperplasia, eosinophil microabscesses, and angioproliferation. The surgical resection (with a safe margin of healthy tissue) with/without initial high-dose steroid therapy is the treatment of choice with risk of recurrence; regression should be monitored over a prolonged period of time.

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Sialectasis of Stensen's Duct With an Extraoral Swelling: A Case Report With Surgical Management

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The definition of sialectasis, according to the Merriam Medical dictionary, is "a dilated salivary duct." When dealing with Stensen's duct, such dilations occur as a

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Dilations will vary in size depending on the severity of the obstruction, the elasticity of the duct, and the degree of gland function. In the case of chronic parotitis, mild to moderate dilations will be encountered, resulting in the so-called "sausage effect" where

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