Kimura's Disease in the Parotid Gland

? Can, V Gündüz, H Pulat, E Samim

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Abstract

Kimura's disease is an immune mediated inflammatory disorder that usually involves the head and neck region, primarily affecting the salivary glands, adjacent muscle and regional lymph nodes. This is usually accompanied by peripheral blood eosinophilia and elevated serum IgE levels. It occurs usually in Orientals with rare cases having been reported in Caucasians. Clinically and radiologically, it is difficult to differentiate Kimura's disease from salivary gland malignancy, lymphoma, lympangioma or haemangioma. The clinical, radiological, surgical and pathological findings of a patient who presented with a right facial mass involving the right parotid gland and the adjacent muscles are discussed in this article.
Kimura disease should be kept in mind in differential diagnosis of parotid gland masses. In the management, surgical treatment should not be so aggressive.

INTRODUCTION

Kimura's disease (KD) is an immune mediated inflammatory disorder that usually involves the head and neck region, primarily affecting the salivary glands, adjacent muscle and regional lymph nodes [1, 2].

The clinical, radiological, surgical and pathological findings of Kimura's Disease were discussed in this article.

CASE REPORT

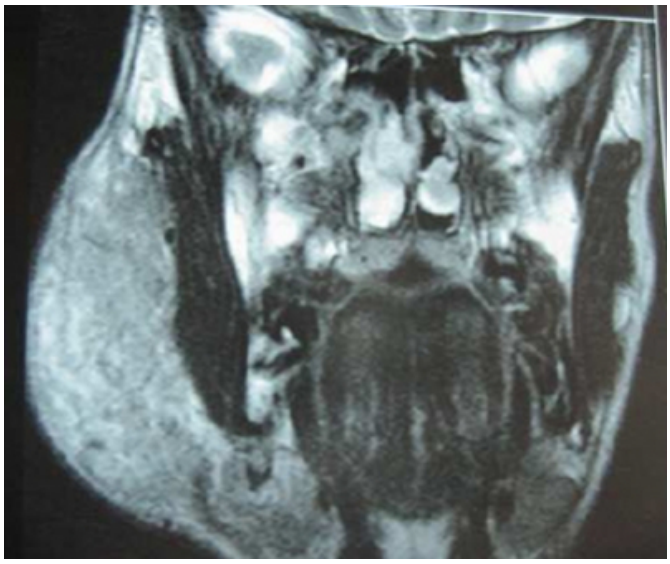
A 21 year old Caucasian man presented with a history of slowly growing, painless right parotid mass. He had a right parotid mass for 4 years. He had used parenteral antibiotics at that time for his right parotid and cheek swelling. He denied any recovery form that treatment as well as weight loss, night sweating, allergy and any other systemic disease. Other history was unremarkable.

On physical examination, he had a 4x8x9 cm cystic, painless mass on his right parotid and cheek region on palpation. The skin over the parotid area was normal. Other findings were unremarkable. All laboratory findings were within normal limits except blood eosinophil count (7%) and total IgE level (3600, normal up to 100).

On ultrasonography (USG), he had a 69x50x86 mm solid, homogenous mass on the right parotid gland with same ecogenity of the gland. On MRI, he had a 4.5x8x9.5 cm subcutaneous mass in relation to parotid gland posteriorly and in relation to surrounding muscles anteromedially (Figure 1).

Figure 1

Figure 1: MRI view of the lesion



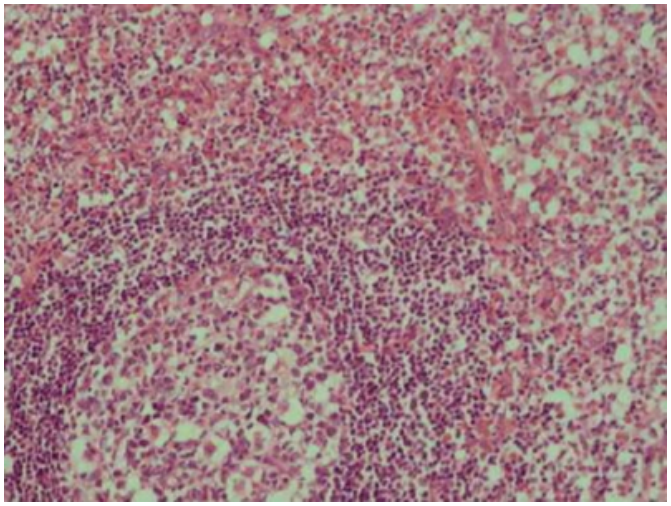
The subcutaneous tissue was obliterated with this mass. He had fine needle aspirations three times reported as benign lymphoepithelial lesions and intraparotideal lymph node hyperplasia (granulomatosis). The preoperative diagnosis was lymphanjioma or hemangioma clinically despite the solid nature of the lesion radiologically.

Right superficial parotidectomy and mass excision were performed on this patient. He had an 8x10 cm mass with unclear borders and one cm lymphadenopaty. A

characteristic appearance was the adherence of the skin to the parotid mass, with no clear plan of dissection between the lesion and the parotid tissue. Identification of the facial nerve was difficult because of the leathery fibrotic appearance of the dissection zone. The mass was not cystic but solid opposing the preoperative palpation findings. Frozen section investigation did not give a clear diagnosis. Final histopathology was reported as Kimura disease showing proliferated lymphoid follicles surrounded by eosinophils and plasma cells in the salivary gland and neighboring cheek tissue. These eosinophils come together forming local abscess areas (Figure 2).

Figure 2

Figure 2: Histopathology of the lesion, H&E 100



He was put on steroid therapy postoperatively starting 60 mg daily and tapering gradually. The mass totally disappeared after this treatment. He does not have any recurrence two years postoperatively.

DISCUSSION

KD is a rare, chronic inflammatory disorder that is almost exclusive to Orientals. Several etiological factors have been accused including autoimmune, allergic and infective causes such as insect bites, parasites and Candida, although no infective agent has been isolated so far in lesions of KD [2, 3]. Allergy may be an etiological factor due to the high IgE and eosinophil levels in the blood in this patient.

Diagnosis of KD is frequently difficult. A biopsy or excision of the lesion is frequently required for definitive diagnosis. The plain radiographic investigations are of no use. Both computed tomography appearance and USG findings are also non specific [4].

Patients with KD and nephrotic syndrome have been reported. That's why these patients should be investigated and followed up for renal disease [5].

Clinical differential diagnoses of KD include lymphoma, lymphangioma, reactive lympadenopathy, salivary gland tumor, nodal metastasis, Mikulicz's disease and angiolymphoid hyperplasia with eosinophylia [2].

The optimal treatment of this disease is not well established. However, the aim should be to preserve the cosmetic and function while preventing recurrences and sequels. Treatment is usually complete surgical excision although recurrences are a problem. The boundary of the granuloma and the surrounding soft tissue is usually unclear, making complete resection difficult. Thus surgery alone cannot control this disease. Other treatment options are oral steroid therapy, intralesional steroid therapy, cytotoxic agents such as cyclosporine and pentoxyphyline, low dose radiotherapy, cryotherapy, pentoxiphyline, nonsteroidal anti-inflammatory drugs, oral retinoid, laser fulguration and surgical excision. The overall prognosis is good, spontaneous resolution can occasionally occur and no malignant transformations have been reported [2].

CONCLUSION

If there is a solid mass in the parotid region with blood eosinophilia and high total IgE, Kimura's Disease should be included in differential diagnosis. If diagnosis can be made preoperatively, steroid therapy may be tried without surgical risk of facial paralysis.

CORRESPONDENCE TO

İlknur Haberal Can Camlıca Bulvar Sitesi F Blok No:19 Umitkoy Ankara Turkey Phone: 905325875338 Email: haberalcan@gmail.com

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Author Information

?lknur Haberal Can, M.D.
Attending physician, ENT Clinic, Ministry of Health Ankara Education and Research Hospital

Veli Gündüz, M.D.
Resident, ENT Clinic, Ministry of Health Ankara Education and Research Hospital

Haluk Pulat, M.D.
Attending physician, Pathology Clinic, Ministry of Health Ankara Education and Research Hospital

Ethem Erdal Samim, M.D.
Attending physician, ENT Clinic, Ministry of Health Ankara Education and Research Hospital