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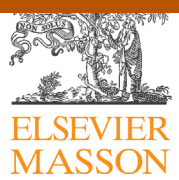

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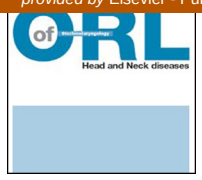

CASE REPORT

Intraparotid Kimura disease

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KEYWORDS
Kimura disease;
Parotid;
MRI;
Hypereosinophilia

Summary
Introduction: Intraparotid locations are extremely rare in Kimura disease, especially in Europe.
Case report: A 31-year-old man presented with intraparotid Kimura disease, managed by parotidectomy.
Discussion/conclusion: The case was analyzed in the light of a review of the literature, focusing on the diagnostic and anatomopathologic problems encountered, and the physiopathology and treatment of this pathology. Any parotid mass found in a patient of Far-Eastern origin showing hypereosinophilia should suggest a diagnosis of intraparotid Kimura disease.

Introduction

Kimura disease, or eosinophilic lymphogranuloma, is a rare condition, first reported in China in 1937 [1] and described anatomopathologically by Kimura [1]. It is endemic in the Far East (China and Japan) [2—4]. Parotid locations are exceptional, with less than 40 cases described in the literature; cases reported in Europe are even rarer.

Case report

Mr. VH., 31-years-old, of Cambodian origin, consulted for a left parotid mass 4 cm in diameter. He had undergone surgery for a mass located in the same region, 10 years previously; no records of this were available. There was no facial palsy or satellite adenopathy. The tumor was mobile and non-inflammatory. Biological analysis found hypereosinophilia (8100 leukocytes, 17.2% eosinophilic) and strongly elevated total IgE.

On MRI (Fig. 1), the tumor signal was heterogeneous and intermediate on T2-weighted images. Apparent diffusion coefficient ratio was elevated 1.6-fold. There were small (subcentimetric) ipsilateral level Ib and V cervical adenopathies, with homogeneous signal. A diagnosis of recurrence of pleomorphic adenoma was considered. Cytologic biopsy was not undertaken, as the patient was severely psychotic, making cooperation difficult.

Left parotidectomy was performed under VII nerve monitoring (4-channel NIM nerve monitor), with wide-margin monobloc resection, conserving the capsule and including the superficial and deep facial lobes. A ring of sternocleidomastoid muscle adhering to the lesion, the facing musculo-aponeurotic system and the cutaneous scar left by the previous operation were resected. The facial nerve and external carotid artery were conserved.

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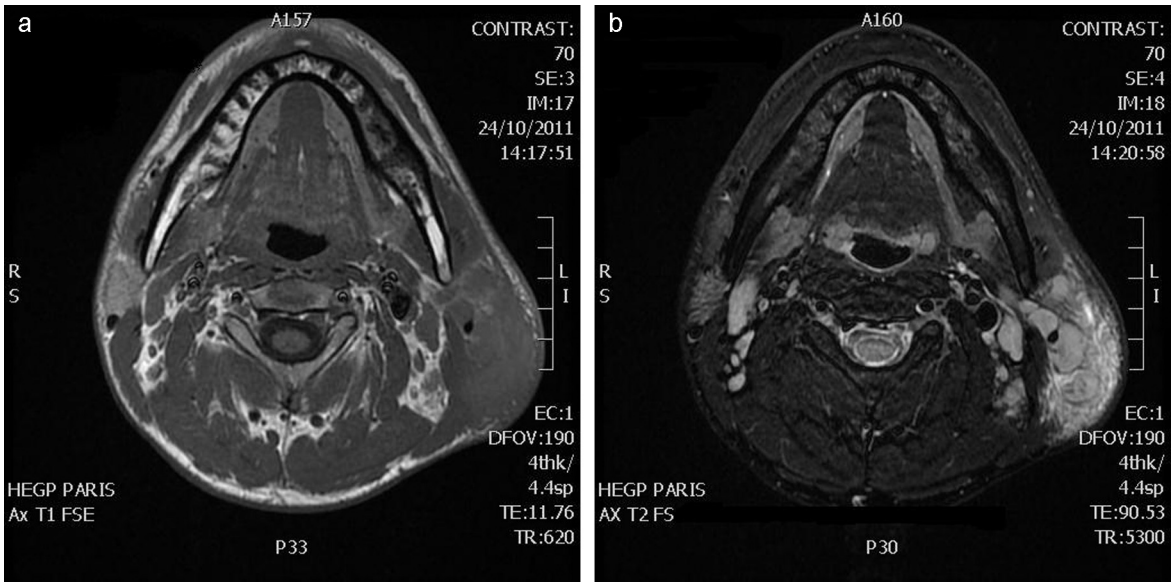

Figure 1 a : T1-weighted sequence ; b : T2-weighted sequence. Pre-operative MRI showing a 4 cm diameter tumoral mass infiltrating subcutaneously and in contact with the skin facing a fibrous scar showing in hyposignal on T2-weighted sequence. The tumor is infiltrative, with heterogeneous intermediate signal on T2-weighted sequence.

Extemporaneous anatomopathologic examination found a benign aspect, possibly with intraparenchymatous nodes. Postoperative course was free of facial palsy or other complications. On macroscopic cross-sectional examination, the parotid showed several more or less well-contoured subcentimetric tan-colored nodules. Histology found the parotid parenchyma partially destroyed by a large follicular mass suggestive of an intraparotid node (Fig. 2). The lesion was separated from the rest of the parotid gland by fibrous trabeculae, though infiltrating the gland here and there, dissociating the glandular structures, surrounding the neural structures and dissociating the striate muscle fibers. The follicular hyperplasia comprised large clear-centered follicles homogenized by a mixed population predominated by small lymphocytes with a few centroblasts. These germinal centers showed dendritic follicular cells, hypertrophic hyperplastic Warthin-Finkeldey cells, and clusters of polynuclear eosinophils (forming little pseudonecrotic clusters) (Fig. 3). The interfollicular areas showed vascular hyperplasia with polynuclear eosinophils. On immunohistochemistry, the lymphoid follicle phenotype was CD20+ CD5− CD10+ BCL2− BCL6+, expressing an elevated mitosis index. Anti-cytokeratin antibodies showed no lympho-epithelial lesion.

Kimura disease was diagnosed.

Discussion

Kimura disease is a rare condition, endemic in the Far East (China and Japan) but exceptional in Europe. Less than 200

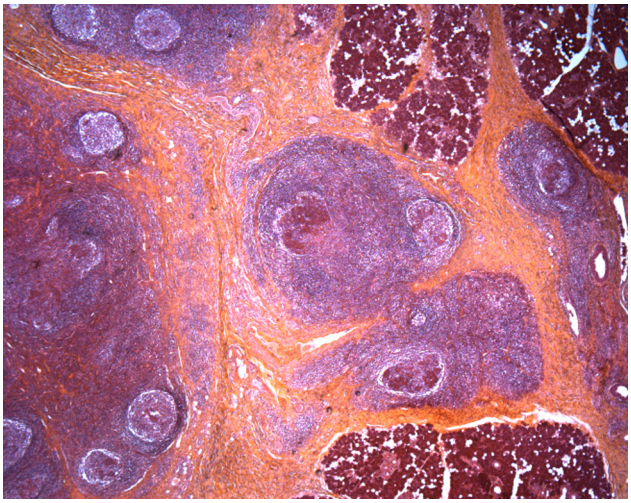

Figure 2 Histologic aspect of parotid parenchyma partially destroyed by a large follicular lymphoid mass (HES × 40).

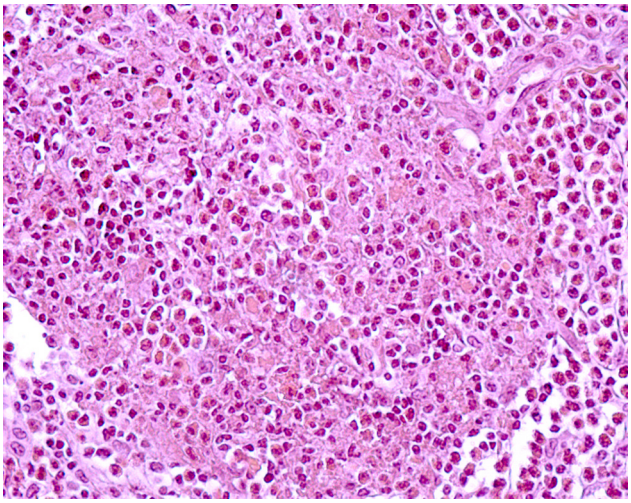

Figure 3 Presence of polynuclear eosinophil infiltrate with pseudonecrotic clusters inside a germinal center (HES × 400).

cases have been reported in the literature, with less than 40 involving a parotid location. Victims are typically males in their 30s: the sex ratio is between 4 and 7 [2—5]. It is an inflammatory pathology of unknown etiology: intense immune reaction to an unknown antigen stimulation has been suggested [5].

Clinically, the disease presents as painless pruriginous cutaneous head and neck nodules, progressively increasing in volume; the skin is not inflammatory [5]. Associated locations have exceptionally been reported: kidney (extramembranous glomerulonephritis), nerve, orbit, spermatic cord [2]. The present patient had no associated lesions. Blood analysis found hypereosinophilia and total IgE elevation.

Salivary gland involvement is rare; most cases are isolated [6—8] and only three series have been published [2—4]. Takeishi et al. [2] reported a series of 11 patients (mean age, 31 years), presenting with temporal frontal skin lesions; two (18%) showed parotid involvement, treated by parotidectomy and radiation therapy, with recurrence in both cases. According to Iwai et al. [3], in a series of nine patients with parotid location, parotid Kimura disease should be considered in case of more than 10% hypereosinophilia associated with a parotid mass, in agreement with the present case. Park et al. [4], in a series of 28 Kimura patients, reported 23 with parotid involvement; on MRI, lesions showed in iso- or slightly hyper-signal on T1-weighted images and intense hyper-signal on T2, slightly enhanced on contrast medium injection; this description matches the present case (Fig. 1a and b), with some atypia attributable to the previous surgical scar.

Anatomopathologic examination of the cutaneous and salivary lesions found florid hyperplasia of the germinal centers (Fig. 2) and polynuclear eosinophil infiltration of the interfollicular compartments (Fig. 3). The closest histologic differential diagnosis would be exceptional angiofollicular hyperplasia with eosinophilia [9] which, however, mainly affects Western females, is not associated with IgE elevation, and is clinically restricted to cutaneous and not parotid involvement.

Treatment is surgical, but recurrence is frequent (as in the present case). Corticosteroids have been recommended in case of renal involvement [8], with possible recurrence at end of treatment. In a few cases, postoperative radiation therapy has been applied, but failed to prevent recurrence [2,10]. The physiopathologic mechanism my lie in TH cell and TH2 cytokine activation inducing B cell activation and eosinophilic infiltration; such an activation cascade suggests that cyclosporine therapy could be appropriate [10].

Conclusion

Kimura disease with parotid location is extremely rare, but should be considered in case of a parotid mass associated with hypereosinophilia and/or strongly elevated IgE levels in Far-Eastern patients.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References

[1] Kimura T, Yoshimura S, Ischikawa E. Unusual granuloma combined with hyperplastic changes in lymphatic tissues. Trans Soc Path Jpn 1948;13:179—80.
[2] Takeishi M, Makino Y, Nishioka H, et al. Kimura disease: diagnostic imaging findings and surgical treatment. J Craniofac Surg 2007;18:1062—7.
[3] Iwai H, Nakae K, Ikeda K, et al. Kimura disease: diagnosis and prognostic factors. Otolaryngol Head Neck Surg 2007;137:306—11.
[4] Park SW, Kim HJ, Sung KJ, et al. Kimura disease: CT and MR imaging findings. AJNR Am J Neuroradiol 2012;33:784—8.
[5] Larroche C, Blétry O. La maladie de Kimura. Encycl Orphanet 2005:1—3, http://orpha.net/data/patho/FR/fr-kimura.pdf
[6] Yazici D, Tuncer U, Ergin M. Kimura disease of the parotid and retroauricular region: a case report. Arch Otolaryngol Head Neck Surg 2007;133:86—9.
[7] Bağdatli D, Kara IG, Bir F. Kimura disease in the parotid gland region. Otolaryngol Head Neck Surg 2008;37:E76—9.
[8] Uysal IO, Eryilmaz MA, Salk L, et al. Kimura disease in the parotid gland. J Craniofac Surg 2011;22:337—8.
[9] Ussmüller J, Donath K, Shimizu M, et al. Differential diagnosis of tumorous space-occupying lesions of the parotid gland: angiolymphoid hyperplasia with eosinophilia and Kimura disease. Laryngorhinootologie 1997;76:110—5.
[10] Sato S, Kawashima H, Kuboshima S, et al. Combined treatment of steroids and cyclosporine in Kimura disease. Pediatrics 2006;118:e921—3.