Kimura’s Disease in Upper Arm: a case report and imaging findings

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Kimura disease (KD) is a chronic benign disorder, primarily seen in Asian males. The major physical manifestation of this disorder is slowly enlarging subcutaneous tumors characterized by the presence of eosinophilic lymphoid hyperplasia [1].

We report a case of Kimura’s disease involving the subcutaneous layer of the left elbow, present with a palpable painless mass enlarging slowly over the past eight months. MRI shows an ill defined subcutaneous mass of high signal intensity on T1- and T2-weighted images with intense enhancement, surrounding subcutaneous edema, and internal flow voids which are typical of Kimura’s disease.

Kimura’s disease is a rare chronic inflammatory disease of unknown origin, primarily seen in young Asian males during the second and third decades [2, 3]. The major physical manifestation is a slowly enlarging subcutaneous mass at the head and neck region, or painless unilateral cervical lymphadenopathy [4-9]. Subcutaneous masses are less commonly seen in axilla, popliteus, groin, and forearm [7, 8]. Other features of Kimura’s disease include peripheral blood eosinophilia, regional lymphadenopathy, and occasional nephrotic syndrome. Here we report a case of Kimura’s disease presenting as a subcutaneous soft tissue mass at left elbow of a 41-year-old Asian male.

**CASE REPORT**

A 41-year-old male patient visited our orthopedic department with the complaint of a palpable painless mass at his left elbow for eight months. Physical examination revealed a soft, non-tender, immobile, subcutaneous mass at the epitrochlear region of left elbow without change in skin color. The patient denied any travel history, medical history or history of contact with cats or other animals.

The laboratory data including complete blood count and electrolyte were all within normal limit, but the eosinophilic count and serum immunoglobulin E level were not obtained.

Radiographs of the left elbow revealed unremarkable bony structure. Then, MRI was performed to evaluate the mass. The MR imaging showed a subcutaneous mass at the level of distal humerus, with ill-defined margin and peripheral soft tissue stranding (Fig. 1). The signal intensity of the mass was slightly higher than or equal to that of muscle on T1 weighted images (Fig. 1a) and relatively higher than that of muscle on T2 weighted images (Fig. 1b). The mass showed enhancement on gadolinium-enhanced images (Fig. 1d, 1e). There were serpentine signal voids in the mass, which indicated neurovascular bundles (Fig. 1a, 1b).

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After excision of the mass, histologically, it showed marked hyperplasia of lymphoid follicles. Diffuse eosinophilia and eosinophilic microabscess were seen in the germinal center. Hyperplasia of small to medium-sized blood vessels was noted. There were also nerve bundles (Fig. 2). The feature was consistent with Kimura’s disease.

**DISCUSSION**

Kimura's disease was first described in China in 1937 by Kim and Szeto [10], but was not widely recognized until a more systemic description by Kimura in the Japanese literature in 1948 [1]. Case reports of Kimura’s disease in the upper extremity have been published sporadically, but there was only one original article focusing on the imaging findings [11].

Our case shows features mostly consistent with Kimura’s disease described by Choi et al. [11] Reviewing the literatures, all the masses showed signal intensity similar to or slightly higher than that of muscle on T1-weighted images and high signal intensity relative to muscle on T2-weighted images. On gadolinium-enhanced T1-weighted images, homogeneous enhancement and signal-void structures within the mass were observed in all cases.

**Figure 1.** a. Axial spine echo T1-weighted MR imaging (TR/TE=663/20) reveals ill defined soft tissue mass (arrows) with iso-signal intensity to muscle in medial subcutaneous tissue of left distal humerus with internal flow voids (open arrow). b, c. Axial and sagittal fast spin-echo STIR imaging (TR/TE/IR=1200/15/165) shows high signal intensity of mass, internal flow void (open arrow), and surrounding soft-tissue infiltration(arrows). d, e. Axial and sagittal spin-echo gadolinium-enhanced T1-weighted MR imaging (TR/TE = 1143/20) with fat suppression shows enhancement of mass (arrows).
In consideration of the imaging findings, the list of differential diagnoses include various inflammatory and neoplastic conditions, such as tuberculous lymphadenopathy, cat scratch disease, lymphoma, metastasis, and primary soft tissue tumor. Lymphadenopathy of tuberculosis tends to have a central low attenuation area and a ring like contrast enhancement [13]. Cat scratch disease is an infectious lymphadenitis frequently occurring in children and adolescents, which was characterized by extensive stranding of the surrounding soft tissues. A patient with cat scratch disease usually has the history of contact with cats or animals. Lymphoma and metastases do not have the characters of extensive soft tissue stranding [12] and internal signal-void structures. Surrounding extensive soft tissue stranding is not often seen in primary soft tissue tumors [12]. A precise analysis of the characteristics of MR findings and clinical information is helpful to make a differentiation [13].

The MRI findings of Kimura's disease are non-specific. The diagnosis is based on histopathology. In our case, the diagnosis of Kimura's disease is also depended on the histopathologic diagnosis. In histopathology, Kimura's disease is characterized by prominent germinal centers in the involved lymph nodes with cellular, vascular and fibrous components, and also with eosinophilic infiltration. We have these findings in our case.

In conclusion, when the imaging findings show a subcutaneous soft tissue mass with iso- to high signal intensity on the T1-weighted images and high signal intensity on the T2-weighted images, intense enhancement on the gadolinium-enhanced T1-weighted images and signal-void structures within the mass, Kimura's disease should be considered, if there are clinical presentations of painless soft-tissue mass, blood eosinophilia and high serum IgE level [13].

REFERENCES

發生於上臂的木村氏病：病例報告及影像特徵

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木村氏病是一種少見慢性良性的疾病，發生於亞洲男性，它主要的特徵是具有嗜酸性淋巴血管增生的腫塊長在皮下組織內，並且緩慢增長。我們報告了一個長在左肘的木村氏病例，這個病例以一個無痛且慢性增長的腫塊來表現。病人接受了核磁共振掃描。文中我們將描述核磁共振的特徵及開刀後的病理特徵。