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Pseudoaneurysmal angiolymphoid hyperplasia with eosinophilia and IgG4–related disease: a possible overlap?

Pseudo-anévrisme secondaire à une hyperplasie angiolymphoïde avec éosinophiles et maladie sclérosante à IgG4: possible forme de chevauchement?

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Conflits d’intérêts : aucun

Ce travail n’a fait l’objet d’aucune subvention.
A 47-year-old right-handed man was referred for the assessment of a growing and pulsating mass of the inner side of the left elbow over the last 6 months. Apart from a venous puncture and playing tennis, he has no previous history of local trauma, autoimmune disease, surgery or smoking. Clinical exam was normal. A Doppler ultrasound, followed by an angio-computed tomography, showed multiple small vessels around the bifurcation of the left brachial to radial and ulnar arteries and without abnormalities of the arterial wall, suspecting a 30 mm pseudoaneurysm (figure 1). Surgery was performed to remove the vascular tumour. The pseudoaneurysm arose from the ulnar artery, involved the radial artery and was adhesive to the surrounding tissues (figure 1). After the excision, arterial reconstruction has included a brachial to ulnar bypass with the left saphenous vein. Histopathological examination revealed a destruction of the arterial wall with an old thrombosis and a dense eosinophilic and lymphoplasmacytic infiltrate. Immunohistochemical staining showed an elevated number of mature IgG4+ plasma cells (CD79a, CD138) (ratio IgG4+/IgG: 50%) (figure 2). Taking together these elements allowed the diagnosis of angiolymphoid hyperplasia with eosinophil (ALHE) associated with IgG4 related disease (IgG4-RD). Normal IgG4 level (63mg/dL) and no antineutrophil cytoplasmic antibody were detected in patient’s serum. $^{18}$F-fluorodeoxyglucose (FDG) positron emission tomography demonstrated an increase FDG activity (SUV max: 9.4) involving bilateral orbital adnexa, without clinical impact after a complete ophthalmological assessment. Thus, no immunosuppressive treatment was added and the patient remained asymptomatic after 3 years.

On one side, ALHE is usually a rare benign vasoproliferative tumor with an inflammatory lymphocytic and eosinophilic infiltration localized on the head and neck. However, three unusual cases of ulnar pseudoaneurysm have been reported so far (1). On the other side, IgG4-RD is a rare polyclonal lymphoplasmacytic infiltration with IgG4 positive plasma cells affecting multiple organs. As IgG4-RD might be associated with hematological conditions,
including Castleman’s disease and lymphoma, differential diagnosis can be difficult. Moreover, extra-aortic lesions of the vascular IgG4-RD are rare and often restricted to the branches of the aorta (iliac, mesenteric arteries) with the impairment of another organ in 71% of cases (2). In our patient, the triggering effect of a local trauma, including a deep venous puncture with an arterial wound, remains uncertain (1). Finally, an overlap between ALHE and IgG4-RD was proposed with unusual features of both diseases because they share similar histological patterns and similar case has already been reported in the literature (3). Thus, a complete clinical, biological and radiological work-up needs to be performed to stage the disease and search for other location, as orbital involvement found in our patient.

References


**Figure 1.** Angio-computed tomography. Pseudoaneurysm measuring 30 mm (A) with multiple small vessels around the bifurcation of the left brachial to radial and ulnar arteries. Surgical view of the excision (B)

**Figure 2.** Histopathological examination. Destruction of the arterial wall with an old thrombosis and a dense eosinophilic and lymphoplasmacytic infiltrate (D). Immunohistochemical staining showed an elevated number of mature IgG4+ plasma cells (CD79a, CD138) (E)