LETTER / ORL

Imaging of inflammatory myofibroblastic cervical tumours: A case report

W. Marraoui,a,*, B. Jeana, M. Muheishb, S. Trouillierc, J.-L. Kemenyd, F. Dorcierb

a Department of Radiology and Medical Imaging, CHU Gabriel-Montpied, 58, boulevard Montalembert, 63003 Clermont-Ferrand cedex 1, France
b Department of Radiology and Medical Imaging, CHR Aurillac, CHR Henri-Mondor, 50, avenue de la République, BP 229, 15002 Aurillac cedex, France
c Department of Internal Medicine, CHR Henri-Mondor, 50, avenue de la République, BP 229, 15002 Aurillac cedex, France
d Department of Pathological Anatomy and Cytology, CHU Gabriel-Montpied, 58, boulevard Montalembert, 63003 Clermont-Ferrand cedex 1, France

KEYWORDS
Inflammatory myofibroblastic tumour; Disseminated erythematous lupus; Imaging; Corticotherapy

Among the family of mesenchymatous tumours, inflammatory myofibroblastic tumours (IMT) are an increasingly recognised and defined lesional group due to the great many studies and recent publications. The aetiopathogenicity is still not fully understood. It seems that an immune origin is involved in the pathological process [1,2]. These masses result from the proliferation of fibroblast and lymphocyte cells associated with collagen wickerwork. Most often benign, they may affect any organ or supporting tissue. However, these lesions remain non-specific and are difficult to distinguish from malignant tumoral processes. Here resides the value of imaging: to make the diagnosis, guide the biopsy and thereby avoid early damaging surgery or an aggressive medical treatment. This is all the more valid since recent papers have shown that corticotherapy of short duration is currently the first intention treatment [2,3]. We here describe the case of a female patient presenting disseminated erythematous lupus who presented a cervical myofibroblastic tumour. The early diagnosis allowed for the initial medical care, which turned out to be effective without resorting to surgery.

* Corresponding author.
E-mail address: Marraouiwissam@yahoo.fr (W. Marraoui).

2211-5684/S — see front matter © 2012 Éditions françaises de radiologie. Published by Elsevier Masson SAS. All rights reserved.
doi:10.1016/j.diii.2012.03.024
Case report

A 51-year-old woman came to the emergency unit for painless left jugal and sub-mandibular tumefaction associated with the installation, over 5 days, of trismus that became irreducible. In the clinical examination, the patient was apyretic and the anamnestic report a past history of disseminated erythematous lupus that was diagnosed many years ago, for which her poor compliance with the corticotherapy is poor. The laboratory tests detected an inflammatory syndrome without infection. An injected cervico-encephalic scan revealed a poorly defined infiltrating mass that was strongly enhanced by the contrast product without washing late after injection. This lesion occupied the left pterygoid-maxillary fossa and extended to the infra-temporal region near the left temporo-mandibular joint without bone lysis (Fig. 1). An MRI completed the description of this lesion presenting an iso-hyposignal T1, a hypersignal T2, and with contrast enhancement after the injection of gadolinium persisting on a late sequence. All this points to a fibrous component. On the other hand, the examination distinguished an heterogeneity of bone signal of inflammatory appearance without bone lysis (Fig. 1). The dysimmune predisposition and the atypical and mildly aggressive radiological characteristic suggest the diagnosis of a subacute inflammatory process rather than something of malignant origin. Since the possibility of an inflammatory myofibroblastic tumour was also mentioned, a sono-guided biopsy was carried out. By histology, the latter confirmed the diagnosis of IMT (Fig. 2). In the extension assessment, a PET-scan helped eliminate other synchronous locations (Fig. 1). In view of the anatomo-pathological results, the first intention treatment consisted of corticotherapy for 1 month. The control by scanner at the end of treatment showed the disappearance of the enhancement of the contrast product (Fig. 3). The other controls after 3 and 6 months and 1 year did not detect any recurrence. During this period, the patient presented good compliance with her basic treatment for her connectivitis.

Discussion

IMT are a sub-group of mesenchymatous lesions that may affect adults and children. The IMT are currently a separate entity defined by clinical, radiological, histopathological

![Figure 1](image-url). Imaging of the inflammatory cervical myofibroblastic tumour. a: cervical TDM with injection of iodine contrast product in axial sections: highly contrasted infiltrating mass (black arrow) occupying the left pterygoid-maxillary fossa and extending to the left infra-temporal region near the left temporo-mandibular joint without bone lysis; b, c: cervical MRI axial sections T1, T2: revealing the mass syndrome in T1 iso-hyposignal (white arrow), hypersignal T2 (tip of white arrow) without peri-lesional oedema resembling a fibrous component. A left cerebellar ischemic sequence should be noted; d, e: cervical MRI axial, coronal sections T1 after injection of gadolinium chelate late after the injection: presence of distinct enhancement of the mass in coronal section (star) and axial section (black tip of the arrow) without washing late after the injection; f: PET-scan: lesional hyperfixation without other synchronous location at a distance (tip of grey arrow).
and molecular criteria [4,5]. They result from the proliferation of fusiform cells on myxoid background and an inflammatory component. The diagnosis is confirmed by the anatomicopathology that detects the myofibroblastic nature of the cells, positive in 70 to 90% of all cases of smooth muscle actina and in 40 to 70% of all cases of desmina and calponina [4,5]. These lesions may affect all parts of the body. They predominate in the lungs and orbits. A cervical location is rare. It develops more often in men. IMT are lesions with a low degree of malignancy and little recurrence and metastasis (under 10%) [4,5]. Clinically, chronic inflammatory tumefaction is described, rarely accompanied by systemic manifestations such as fever, weight loss, anaemia, hypergammaglobulinemia or an increase in the sedimentation rate [6]. The pathogenicity is still not fully clear although a dysimmune context seems to be a predisposing factor (connectivitis, infection by Epstein Barr virus or herpes 8) [1,2]. As far as we are aware, only was case has been reported in the literature, representing the second case of IMT associated with disseminated erythematous lupus [7]. Imaging is included in all stages of the care of this lesion: descriptive stage (locoregional extension), analytical stage (suggestion of the differential diagnoses) and interventional stage (guiding of the biopsy).

Figure 2. Histo-anapathological sections of the sample taken by sono-guided biopsy: a: HES (× 20): Inflammatory lymphocytary infiltrate in the collagen tissue; b: HES (× 20): extension of the connective tissue to the striated muscle fibres; c: immunohistochemistry with actin: labelling of fusiform cells.

Figure 3. Control after 1 month: cervical scan with injection of iodine contrast product. Absence of contrast within the mass attesting to a good response to the medical treatment (tip of black arrow).
These lesions remain non-specific and may resemble lymphomatous lesions, nasopharyngeal carcinoma or Wegener’s granulomatosis [2]. Nevertheless, certain characteristics orient the diagnosis to cervical locations: the topography, rather at the level of the infra-temporal fossa, the pterygopalatin space and opposite the temporo-mandibular joint. The scan is non-specific and detects a homogenous or heterogeneous hypo, iso or hyperdense lesion taking up the contrast that persists long after injection. The MRI presents more specific criteria: an hypointense T1 and T2 signal associated with distinct enhancement persisting late after the injection calling to mind a fibrous component, possibly accompanied by inflammation of the opposite bone structures without lysis such as bone signal heterogeneity [1,8–10]. These non-specific characteristics make an aggressive process less certain and rather indicate a chronic inflammatory phenomenon. The PET-scan contributes to the assessment of extension in the search for other synchronous locations. Mention of this diagnosis and recognition of these lesions during the pre-surgical period are fundamental. This avoids immediate damaging surgery that is often not indicated. This agrees with the recent papers demonstrating spontaneous regressions or regressions with medical treatment. Moreover, corticotherapy is currently recognised as the first intention treatment after histological proof, necessary in pre-therapy due to the existence of differential diagnoses [2,3]. The most aggressive cortico-sensitive lesions may resemble IMT, first of all the lymphomas.

**Conclusion**

IMT are rare lesions, especially at the cervical level. They may resemble malignant tumours although certain specific features in imaging help point out the diagnosis and thereby orient care towards a less aggressive approach.

**Disclosure of interest**

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

**References**