CASE STUDY

Inflammatory Pseudotumor of the Tonsil

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Abstract Inflammatory pseudotumour is an unusual benign but potentially invasive lesion. In the head and neck zone, it mainly involves the orbit and paranasal sinuses. We present a case in tonsil and neck. After tonsillectomy and steroid therapy, there has been no recurrence.
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Introduction

Inflammatory pseudotumour is a poorly studied entity of controversial histogenesis. Its pathogenesis seems to be an exaggerated response to tissue aggression of unknown aetiology. It poses a complex diagnosis, usually of exclusion, and can appear in any organ. It must be differentiated from lymphoproliferative disorders and low grade spindle cell sarcomas, requiring immunohistochemistry techniques.

Its symptoms depend on its location, although it generally causes displacement, obstruction or invasion. Initially described in the lung, when it appears in the head and neck region, it mainly affects the orbit and paranasal sinuses. Tonsillar location is extremely rare.

Clinical Case

We present the case of a 50-year-old female patient with no relevant medical history who attended consultation due to high left odynophagia, moderate trismus, moderate fever and reflect otalgia with an evolution of 3 months.

Cervical palpation was painful, with an indurated left side without significant lymphadenopathy. The oral cavity presented a hyperplastic and hyperaemic left palatine...
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tonsil, clearly asymmetric, with velar oedema. Nasofibroscopy revealed a permeable cavum and left mobile arytenoid thickening.

Analytical tests revealed leukocytosis with neutrophilia, thrombocytosis, hypochromic microcytic anaemia and elevations in acute phase reactants (APR) and ESR.

The CT scan showed tonsillar hyperplasia and the MRI scan revealed nodular thickening in the nasopharynx and supraglottis with contrast uptake (Fig. 1).

In the absence of clinical response to treatment with antibiotics and steroids, we opted for unilateral tonsillectomy with exploratory endolaryngeal microsurgery and cervicotomy. The arytenoid biopsies were non-specific. The dissection of cervicotomy proved difficult due to intense fibrosis.

Histological examination detected a diffuse, heterogeneous infiltrate of lymphocytes, plasma cells, fibroblasts and neutrophils in the tonsil and cervical fascias, with vascular hyperplasia and desmoplastic reaction of the adjacent stroma, without necrosis. Immunohistochemistry found polyclonality for kappa and lambda light cytoplasmic chains and diffuse, weak positivity against ALK-1, compatible with inflammatory pseudotumour (Fig. 2).

After surgery, the patient improved rapidly. Explorations are currently normal. The corticosteroid therapy was maintained for 6 months.

Discussion

The rarity of inflammatory pseudotumour is not conducive to its initial suspicion in inflammatory processes. It is characterised by presenting an unknown multicellular lineage, a reactive nature and regional aggressiveness.

It was initially described in the lung by Brunn in 1939. The series by Ramachandra et al. with 18 cases, found 33.3% in the head and neck region (orbit and paranasal sinuses), although there have been documented cases with laryngo-tracheal, salivary, middle ear, ganglion, pterygomaxillary and parapharyngeal locations. In 2002, Escobar Sanz-Dranguet et al. presented a case located in the nasal fossae.

Only once previously had a case of tonsillar involvement been described. The torpid evolution in our case indicated tonsillectomy for its histopathological study.

Figure 1  Axial CT sections showing a left tonsillar mass that uptakes contrast and a coronal T2-weighted MRI image showing a left tonsillar mass, abscess and nodules in the veil and pyriform sinus, aryepiglottic fold and parapharyngeal space.

Figure 2  40× image of the tonsil tissue stained with haematoxylin-eosin showing lymphocytes, plasmocytes, neutrophils and fibroblasts (A), immunohistochemical polyclonality for kappa (B) and lambda (C) chains, and weaker granular intracytoplasmic staining against ALK-1 (D).
Such lesions are often unique and simulate neoplasms, chronic inflammatory diseases or largely evolutionary infections, but the cytology, serology and cultures are ineffective. Prior to ours, only 2 cases of ganglionar inflammatory pseudotumour, documented by Moran et al. 6 and Babar-Craig et al. 7 had been multifocal in the neck region.

Biopsy is the procedure of choice for diagnosis, with fusion of cell lines and immunohistochemical characterisation of ALK gene rearrangements being the basis of its typing. 2

No protocols have been established for treatment. Surgical excision is the technique of choice. Corticosteroids offer good results when the lesions are not eminently fibrotic. 1,8 Radiotherapy is a good alternative for conditions with excessively mutilating resections, offering a good response in up to 85% of cases. 9

In our case, tonsillectomy markedly improved the symptoms of the patient. The nodules identified by MRI disappeared and leukocytosis and APR became normalised during the 6 months of corticosteroid therapy.

Conflict of Interests
The authors have no conflicts of interest to declare.

References