CASE STUDY

Inflammatory Myofibroblastic Tumour of the Tonsil: Case Report and Literature Review

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KEYWORDS
Inflammatory myofibroblastic tumour; Inflammatory pseudotumor; Head and neck; Tonsil; Paediatric

Abstract Inflammatory myofibroblastic tumour (inflammatory pseudotumor) is an idiopathic lesion, rare in the head and neck, of unknown aetiology. It is primarily a soft tissue, lung and orbital condition. In the world literature, only two cases with tonsillar disease have been found.

We report a case of a 10-year-old girl admitted to our hospital with clinical complaints of pain in the neck region, cough with vomiting, dyspnoea and dyslexia. Clinical examination revealed halitosis and a neoformation dependent on the left tonsil. Bilateral tonsillectomy was performed.

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Tumor miofibroblástico inflamatorio de amigdala: informe de un caso con revisión de la literatura

Resumen El tumor miofibroblástico inflamatorio (seudotumor inflamatorio) es una lesión idiopática, poco frecuente en cabeza y cuello, de etiología desconocida, que se presenta más comúnmente en tejidos blandos, pulmón y órbita. En la literatura mundial solo se tiene conocimiento previo de 2 casos con afección amigdalaria.

Informamos del caso de una niña de 10 años, que ingresó en nuestro hospital con cuadro clínico de dolor en la región cervical, tos con vómito, disnea y dislexia, la exploración física mostró halitosis y una neoformación dependiente de la amigdala izquierda, se le realizó amigdalectomía bilateral.

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Introduction

Inflammatory myofibroblastic tumour (IMT) is a rare lesion which primarily affects soft tissue, the lungs and orbits. It was first described in 1905 by Birch–Hirschfeld in a patient with unilateral proptosis1 and subsequently designated as inflammatory pseudotumor by Umiker in 1954. 2 Its location...
in the head and neck (extraorbital) is exceptional, with isolated cases being described in the skull base, parapharyngeal space, paranasal sinuses, pterygopalatine fossa and oral cavity.\textsuperscript{3,4} The specialised literature only contains 2 previous cases reported in the palatine tonsil.\textsuperscript{1,5}

Clinical Case

We present the case of a girl aged 10 years and 11 months, with a sudden onset of cervical pain and arthralgia. She was treated with paracetamol, which partially improved her symptoms. Subsequently, she suffered cough and vomiting of gastric contents with traces of blood, as well as foreign body sensation in the throat which limited speech, so she was referred to our hospital. Physical examination revealed a good general condition and the presence of a tumour with irregular edges in the oropharyngeal area. This tumour was dependent on the left tonsil and displaced the uvula, causing pain when touched. The contralateral tonsil showed no abnormalities and the rest of the physical examination was normal. She underwent flexible nasofibrolaryngoscopy, which revealed a grade I adenoid hyperplasia. In addition, we observed that the lesion was in contact with the posterior pharyngeal wall. We performed bilateral tonsillectomy and the pathological findings were as follows: cylindrical left amygdala, yellow-grey in colour, coated with fibrin, measuring 5.2 cm\(\times\)2.4 cm\(\times\)1.5 cm and with a firm and smooth consistency (Fig. 1). Histologically, we observed granulation tissue on the surface, with partial absence of epithelial lining. Within the tumour we observed a disorderly proliferation of dense collagenous bands with stellar and tapered myofibroblasts, as well as a polymorphic, inflammatory infiltrate composed of lymphocytes, plasma cells, mast cells, neutrophils and eosinophils (Fig. 2A and B). Immunohistochemical stains of the stromal cells were positive for vimentin, CD34 and muscle-specific actin, revealing myofibroblastic differentiation, as well as for CD3, CD20, CD68 and CD21, which confirmed the polyclonal nature of the inflammatory infiltrate (Fig. 2C and D). With this evidence, we reported a diagnosis of inflammatory myofibroblastic tumour. We also conducted an ALK1 test, which was negative.

Discussion

IMT has been recently classified as a neoplastic process with myofibroblastic differentiation, accompanied by dense and polymorphic inflammatory infiltrate.\textsuperscript{6} The histological image of inflammatory myofibroblastic tumour is variable and because of this it has had several names throughout history, such as inflammatory pseudotumor or plasma cell granuloma, among others. It mainly affects soft tissues and the lungs.\textsuperscript{7} Its location in the head and neck region (extraorbital) is very rare. There have been reports of cases located in the skull base, parapharyngeal space, paranasal sinuses, pterygopalatine fossa, oral cavity and parotid.\textsuperscript{3,4,8}

At the moment, treatment for IMT is controversial. Throughout history, several authors have proposed therapies based on steroids, excisional biopsy with wide resection, radiotherapy and even chemotherapy, although these treatment models have been described in various locations.\textsuperscript{6} The medical literature only contains 2 reported cases of IMT located in the palatine tonsil. In 1984, Weilbaecher published the case of a 63-year-old man, who was asymptomatic despite presenting an enlarged, right palatine tonsil. This had a firm consistency, measured 3 cm\(\times\)2.5 cm\(\times\)2 cm and was reported as a plasma cell granuloma.\textsuperscript{5} One decade later, in 1995, Newman reported the case of a 62-year-old woman who presented odynophagia as a main symptom, along with an enlarged, left palatine tonsil. This had a firm

Figure 1  Macroscopic appearance of the outer surface (left) and section (right).
consistency, an erythematous appearance and measured 3.5 cm × 2.5 cm × 2 cm. It was reported as an inflammatory pseudotumor.¹

In conclusion, we present a case of primary IMT in the palatine tonsil, which represents the third case described in the literature and the first in a paediatric patient. Our patient presented enhanced symptoms compared with previous cases, probably owing to the fact that this tumour was the largest one reported so far. From a clinical standpoint, it is important to be aware that IMT can affect the palatine tonsil, so it should be included in the differential diagnosis of primary lesions which appear with unilateral tonsillar hypertrophy. Unilateral tonsillectomy is controversial in such cases, but we believe that it should be the treatment of choice.

Conflict of Interests

The authors have no conflicts of interest to declare.

References