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Case Report

Inflammatory Myofibroblastic Tumor of Maxillary Sinus: A Case Report

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Abstract

Inflammatory myofibroblastic tumor (IMT) is an immunohistochemically diverse condition with neoplastic and non-neoplastic characteristics. It is an uncommon tumor that can occur anywhere in the body. Most cases occur in the lung and abdomen and present benign behavior. When located in the nasal cavity, paranasal sinuses, and pterygopalatine fossa, it has increased neoplastic and invasive potential. IMT in the head and neck region is very rare, with few cases described in the literature.

The current case involves a 66-year-old patient with an IMT presenting aggressive behavior in the right maxillary sinus.

Keywords: Inflammatory Myofibroblastic Tumor; IMT; Nasal Cavity; Paranasal Sinuses

Introduction

Inflammatory myofibroblastic tumor (IMT) is an uncommon neoplasm. Most cases occur in the lung and abdomen and behave as benign lesions. First described in 1939 by H. Brunn, IMT is an immunohistochemically diverse tumor and demonstrates neoplastic and non-neoplastic features. Because of similar histological features, IMT was previously classified as an inflammatory pseudotumor. Lesions are histologically described as containing varying degrees of spindle cell proliferation under a myxoid/collagenous stroma and an inflammatory infiltrate composed of lymphocytes, histiocytes, plasmocytes, neutrophils, and eosinophils. Lesions located in the nasal cavity, paranasal sinuses, and pterygopalatine fossa have increased neoplastic and invasive potential when compared to IMT in other locations. Although IMT can occur anywhere in the body, it is rarely located in the nasal cavity and paranasal sinuses. Only about 40 cases have been described in the literature [3].

Case Report

SMRC is female, 66 years old, Brazilian, housewife, taking Trazodone for depression. She went to the otorhinolaryngology outpatient clinic at Madre Teresa Hospital, Belo Horizonte, Brazil due to pulsatile tinnitus symptom and right ear effusion.

Patient also reported chronic right nasal obstruction and facial pain, which were unsuccessfully treated previously as rhinosinusitis.

She presented good nutritional health, without loss of weight or muscle mass. Facial mimic was symmetrical, and extrinsic ocular movement was normal. No palpable masses or changes in the cervical region were found. Otorhinolaryngological examination revealed effusion in the right ear, V2 hypoesthesia, bulging of the hard palate, and a lesion in the right nasal cavity. The lesion presented firm consistency, irregular surface, pale color, and occupied the entire middle meatus and right nasal cavity.

The data compiled were sufficient for clinical consideration of expansive lesion of right nasal cavity and paranasal sinuses. A video nasopharyngoscopy exam identified the presence of a bulging lesion in the right middle meatus, occupying the nasal fossa and impeding the progression of the fiber optic nasopharyngoscope. Initial biopsies showed only sinonasal mucosa with chronic inflammation.

Computed tomography (CT) and magnetic resonance imaging (MRI) of the sinuses revealed an extensive and destructive mass in the right maxillary sinus, extending to the palate, pterygopalatine fossa (PPF), pterygoid musculature, and infratemporal fossa (ITF) (Figures 1a and 1b).

Figure 1: Axial (a) and Sagittal CT (b) views of sinus showing right maxillary sinus mass.

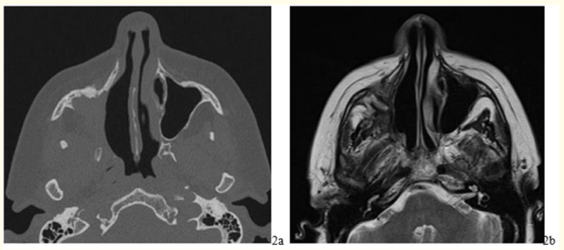

Figure 2: Axial CT (a) and MRI (b) views one year after surgery, demonstrating no evidence of disease.

For diagnostic clarity and treatment, the mass was excised using a combination of two techniques; endoscopic nasal surgery and Caldwell Luc. Using an endoscopic approach, the medial and posterior walls of the maxillary sinus were removed. The PPF was reached, and most of the tumor was excised. As complete resection was impossible using only the nasal approach, Caldwell Luc was also used to access the ITF through the maxillary sinus and completely remove the tumor.

Anatomopathological examination revealed the mass consisted predominantly of "spindle cells with discrete atypia, elongate nuclei, delicate chromatin, scarce cytoplasm, and rare mitotic figures. From the inside we observed some mast cells and a small number of mature lymphocytes without atypia. There are still outbreaks of hemorrhage and mild myxoid edema". Immunohistochemistry test showed fusocellular neoplasia with elongated or more epithelioid cells and eosinophilic cytoplasm. The stroma contained alternating areas of collagenic stroma or more myxoid edema. In the immunohistochemical evaluation, there was isolated expression of smooth muscle actin (Figure 3), suggesting myofibroblastic myoid differentiation. The neural and epithelial differentiation markers were negative. No sarcomatous features were present. The S100, desmin, P63, myogenin, cytokeratin, and CD34 markers were negative. In conclusion, the findings were a spindle cell mesenchymal neoplasm, without clearly defined histogenesis and biological behavior.

Figure 3: Immunohistochemistry, positive neoplasm for smooth muscle actin.

According to the exams, the pathology was consistent with inflammatory myofibroblastic tumor of the maxillary sinus. The patient evolved well postoperatively, without recurrence of tumor after two year follow-up. The patient only developed two minor postoperative complications, permanent V2 hypesthesia and transient trismus. No additional treatment such as chemotherapy or radiation therapy was used [1-4].

Discussion

IMT is a neoplasm with difficult histological and immunohistochemical diagnosis. The mechanism involved in its etiology is not yet fully understood. However, it is believed to be multifactorial, involving chromosomal aberrations and chronic inflammation.

Since the first cases described in the literature, IMT has drawn attention from pathologists, surgeons, and oncologists due to idiosyncrasy in its biological, immunohistochemical, and therapeutic response. The tumor has been classified in the past as a benign lesion. Both the literature and the present case demonstrate that its behavior may be aggressive and even fatal.

The current gold standard treatment of IMT consists of complete surgical excision and corticoid therapy. However, in refractory cases or in inoperable tumors, new treatments with chemotherapy, radiotherapy, NSAIDs, and COX inhibitors have also been used to achieve pathology remission. Despite positive therapeutic response in some cases of maxillary sinus IMT, subgroups of the tumor at this location remain resistant to all forms of therapy.

Different immunohistochemical markers have been identified to aid in the diagnosis of IMT. When absent, the anaplastic limphoma kinase marker, ALK-1, has predicted more aggressive tumors, possibility of metastasis and recurrence, all which indicate the need for more aggressive treatment [3].

Conclusion

By using the endoscopic endonasal approach for the PPF, the authors avoided patient facial scaring, deformity, and dysfunction. By combining this approach with Caldwell Luc, the authors also achieved a high efficacy of resection without recurrence. The positive results therefore support using this combined approach to safely and effectively manage this rare type of tumor.

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