Inflammatory myofibroblastic tumor (IMT) is a benign lesion composed of myofibroblasts accompanied by varying numbers of inflammatory cells. Various pathogenetic factors have been proposed, but the etiology of most IMTs remains unknown. This article presents a case of IMT occurring in the left maxillary sinus. A 24-year-old man complained of throbbing pain in the maxillary left molars and swelling of the left cheek. His maxillary left second molar was diagnosed as pulp necrosis and root canal treatment performed. After that, his symptoms continued and he was referred to the Department of Otolaryngology. Computerized tomography disclosed compact soft tissue masses in the left maxillary sinus with obstruction of maxillary ostium. Under general anesthesia, the lesions were fully excised. Histopathologically, the lesions were composed of plump or spindled myofibroblasts. Cells were immunoreactive for smooth muscle actin and β-catenin, and were negative for ALK1, CD34, and EMA. The diagnosis was IMT of left maxillary sinus. Although it is very rare, IMT should be included as a differential diagnosis in patients with compact masses in maxillary sinus. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;112:684-687)

Sinus mucosal pain is common source that can mimic toothache. typical of deep visceral tissues, sinus mucosal pain can induce central excitatory effects such as secondary hyperalgesia, referral of pain, and autonomic changes. It is this tendency that gives sinus pain the ability to masquerade as toothache. The close anatomic relationship between the maxillary sinus and the maxillary teeth is well known. As a result, irritation of the maxillary sinus may cause dental symptoms. Conversely, sinusitis may have a dental origin, most often from pulp necrosis and chronic apical periodontitis.

Inflammatory myofibroblastic tumor (IMT) is an uncommon, benign lesion characterized by a mesenchymal proliferation and inflammatory cell infiltration composed of lymphocytes and plasma cells. Although various pathogenic factors have been implicated, the etiology of most IMTs remains controversial. IMT occurs most commonly in the lungs where it presents a genuine benign behavior. Its occurrence in the head and neck area, including mouth and maxillary sinus, has also been reported and is characterized by more aggressive behavior.

The present case report presents an unusual case of IMT in maxillary sinus related to pulp necrosis. Ultrastructural and immunohistochemical inspection of the resected tissue was conducted for confirming the diagnosis and for elucidating the potential of a neoplastic pathogenesis.

CASE REPORT

A 24-year-old man presented to the Department of Conservative Dentistry and Endodontics with throbbing pain in the maxillary left molars and swelling of the left cheek. In his dental history, his left maxillary molars (teeth #12-14) had received root canal treatment and been restored with crowns greater than approximately 3 years before. He had suffered from intermittent dull pain over the left maxillary area after that time. Clinical examinations, negative response to thermal pulp tester (Frigi-Dent; Ellman International, Oceanside, NY) and Electric pulp tester (Digitest; Parkell, Farmingdale, NY) of maxillary left second molar (tooth #15) was detected. The preoperative radiograph showed deep gold restoration and secondary dental caries (Fig. 1, A and B). Pulpal necrosis and symptomatic apical periodontitis was diagnosed, and conventional nonsurgical root canal treatment was planned. After emergency access opening, pulp extirpation was performed. Pulp tissues were totally necrotic, and there was no exudate or pus from the canals.

After 4 days, he returned with left facial reswelling, eyeball pain, and abrupt weight loss. He said that swelling had subsided after the emergency treatment and reswelling began after that day. Positive response to percussion test on tooth #15 and tenderness to palpation on the apical area was detected. After working lengths were determined by electronic apex locator (Root ZX; Morita, Tokyo, Japan) and periapical x-ray (Fig. 1, C), all the canals were enlarged using rotary instruments. The root canals were dried and calcium hydroxide applied as an intracanal medicament. An urgent computer-aided tomographic (CT) scan revealed a compact soft tissue density in the left maxillary sinus with obstruction of maxil-
lary ostium (Fig. 2, A) and focal soft tissue density in the left ethmoid sinus (Fig. 2, B). The patient was referred to the Department of Otolaryngology (ENT), and medication therapy (antibiotics, mucolytics, nonsteroidal antiinflammatory drugs) was performed for 2 weeks.

After 2 weeks, his clinical symptoms were improved, but compact soft tissue masses occupying the left maxillary sinus remained in CT. He was advised to have the operation by ENT surgeon, and the operation for resection of masses occupying the left maxillary sinus was scheduled. In our clinic, root canals were filled with the continuous wave technique using System B and Obtura II, and the tooth was restored with a resin core before the operation (Fig. 1, D).

The excision surgery was performed under general anesthesia. The antral polypoid mass was resected, and partially eroded lateral and superior wall was observed in the left maxillary sinus.

Tissues from the lesions were fixed in 10% formalin and processed for paraffin embedding. Tissue blocks were sectioned for haematoxylin-eosin staining and immunohistochemistry. The histologic appearance of the lesion showed a tumor consisting of plump or spindled myofibroblasts in an edematous myxoid back ground with many blood vessels and an infiltrate of chronic inflammatory cells (lymphocytes, plasma cells; Fig. 3, A). Immunohistochemically, the tumor cells exhibited strong cytoplasmic positivity for smooth muscle actin and β-catenin (Fig. 3, B) and negativity for ALK1, CD34, and EMA. On the basis of the histopathologic and immunohistochemical findings, a diagnosis of IMT was made, and there was no evidence of malignancy.

One month after surgery, tooth #15 was restored with a gold crown. The patient has been regularly followed every 3 months with magnetic resonance imaging (Fig. 4). At the time of writing, he had been free from disease for 15 months.

DISCUSSION

IMTs are chronic inflammatory lesions of unknown origin. They have no common identical cause, although some authors have assumed that any inflammatory stimulus may originate these pseudotumors. The name is given because these lesions simulate neoplasm, both clinically and radiologically.

Although the lesion was established as a single entity in 1994 by the World Health Organization classification, because of its diversity IMT has been defined with various terms: plasma cell granuloma, inflammatory pseudotumor, benign myofibromatoma, inflammatory fibrosarcoma, histiocytoma, and xanthogranuloma. This illustrates its variations in histologic appearance and suggests that the term IMT does not represent a single entity but a group of lesions demonstrating non-specific chronic inflammatory changes.

IMT of the maxillary sinus presents a variety of symptoms depending on the site of origin. The most frequent clinical picture is a nonspecific nasosinusal tumor that grows to a stable state over months or years. It may be associated with facial pain, fever, weight loss, nasal obstruction, nose bleeding, proptosis, increased
lymph nodes, and bone erosion and destruction.\textsuperscript{10,16} Our patient showed an acute course and was eventually cured by surgical resection. Surgical resection is generally considered to be the treatment of choice,\textsuperscript{20} but radiation therapy is also recommended for unresectable and recurrent cases.\textsuperscript{21,22} Several authors advocate corticosteroid treatment based on their experience.\textsuperscript{23,24}

IMTs are characterized by 3 basic histologic patterns:\textsuperscript{25} a myxoid/vascular pattern, a compact spindle cell pattern, and a hypocellular fibromatosis-like pattern. The pathologic examination of our specimen showed edematous myxoid background with many blood vessels and an infiltrate of chronic inflammatory cells. IMT in this case was subcategorized as a myxoid/vascular pattern.

The exact etiology and pathogenesis of these lesions remain unknown. Histologically, they are benign but tend to be locally aggressive and clinically or radiologically mimic malignant tumors. IMT should be considered an intermediate tumor with weaker potential to recur and metastasize than other sarcoma.\textsuperscript{25}

The etiology of IMT is uncertain, but it is considered to be an immunologic host reaction to some inciting agents, such as microorganism, adjacent necrotic tissue, neoplasm, foreign bodies and some types of tissue injury.\textsuperscript{18} In the present case, pulp necrosis on tooth #15 (and maybe #12-#14) can be thought to be the origin of IMT. His old gold restoration had been placed for 3 years, and his symptoms on mastication had happened occasionally after that. Therefore, we could assume that infected pulp tissue had promoted chronic inflammation of maxillary sinus. Conversely, an unknown reason or left maxillary pathology had caused the IMT first, leading to pulp necrosis of tooth #15, or perhaps the

Fig. 2. Computerized tomographic (CT) scan disclosing compact soft tissue masses in the left maxillary sinus with obstruction of maxillary ostium. A, Coronal CT scan. B, Axial CT scan.

Fig. 3. A, Inflammatory myofibroblastic tumor (IMT) is composed of plump myofibroblasts and moderate lymphocyte and plasma cell infiltration (hematoxylin-eosin stain, magnification $\times 100$). B, Immunoreactivity of the IMT cells for smooth muscle actin (magnification $\times 100$).
REFERENCES


