Inflammatory Pseudotumor of the Sinonasal Tract

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Inflammatory pseudotumors (IPTs) are a clinically and histologically diverse group of lesions characterized by a tumor mass of acute and chronic inflammatory cells with a variable fibrous response. IPTs most commonly involve the lung and orbit, but rarely the sinonasal tract. We report a 68-year-old male with an IPT of the sinonasal tract presenting as nasal obstruction and postnasal dripping for several years. A gray–white soft mass was noted in the right nasal cavity. Computed tomography revealed a solid mass filling the right nasal cavity and maxillary sinus. Caldwell-Luc operation with ethmoidectomy (right) was conducted to resect the mass en bloc. Pathology revealed admixture of plasma cells, lymphocytes and eosinophils, confirming the diagnosis of IPT. The patient remained symptom-free over 4 years of follow-up. Awareness of the clinical presentations, histopathologic features and treatment of choice of this rare disease entity is necessary to distinguish it from malignancy and avoid unnecessary management. [J Formos Med Assoc 2007;106(2):165–168]

Key Words: inflammatory pseudotumor, maxillary sinus, nasal cavity, plasma cell granuloma, sinonasal tract

Inflammatory pseudotumor (IPT) is a lesion of unknown etiology that was first described in the lung of two patients in 1939.1 Since then, similar lesions have been reported in almost all parts of the body. IPTs are rarely encountered in the head and neck region, but when they occur, the orbit, larynx, oral cavity, oropharynx, and paranasal sinuses are the most prominent sites for these lesions.2–4 The term pseudotumor was coined because these lesions mimic expansile, invasive malignant tumors, both clinically and radiologically. Therefore, it is important to recognize the distinguishing features of IPTs to avoid unnecessary surgery or radiotherapy. Although IPT of the sinonasal tract is rare, rhinologists must be familiar with its clinical presentation, radiologic features, prognosis and therapeutic management. Here, we report a case of IPT of the sinonasal tract.

Case Report

A 68-year-old male presented with nasal obstruction and posterior nasal dripping, with onset several years ago and progressively worsening. The patient's medical history was otherwise unremarkable except for allergic rhinitis. Physical examination revealed a gray–white mass in the right nasal cavity. Sinoscopy revealed a loose soft tissue mass that completely filled the right maxillary sinus. No cervical lymphadenopathy was detected. Routine laboratory parameters (plain chest radiography, full blood count, erythrocyte

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sedimentation rate, biochemistry and coagulation screen) were normal. Computed tomography (CT) of the nose and paranasal sinuses showed a solid mass filling the right nasal cavity and maxillary sinus with linear enhancement (Figure 1). Chronic paranasal sinusitis with polyposis was suspected.

Caldwell-Luc operation and ethmoidectomy were performed under general anesthesia and the mass was removed en bloc. No visible bony destruction was found intraoperatively. The resected mass was fixed in formalin and sent for histopathologic examination.

Grossly, the mass measured $4 \times 1.5$ cm. The cut surface was homogeneous and gray–white. Microscopic examination showed that the great majority of inflammatory cells were plasma cells. The remainder was a variable admixture of lymphocytes and eosinophils (Figure 2). No neoplastic cells were observed. IPT was diagnosed.

Bacteriologic and mycologic tests of the resected tissue were negative. Serum electrophoresis did not reveal a monoclonal band and the test for Bence-Jones proteins in urine was also negative.

The patient remained free of sinonasal symptoms over 4 years of follow-up.

Discussion

IPTs are rare, benign and slow-growing tumors consisting of inflammatory cells, which can arise in almost all organs. The pathogenic mechanism of IPT is unknown, although various theories have been proposed as triggers for their development, such as unrecognized microorganisms, minor trauma, surgery, and association with other malignancies. Williams et al considered that a localized derangement in the immune response may be an underlying mechanism.Various terminology has been used to describe this non-neoplastic lesion, such as plasma cell granuloma, xanthomatous pseudotumor, and post-inflammatory myofibroblastic tumor. These descriptions illustrate the variations in histologic appearance and suggest that IPT is not a single entity but an umbrella term for any nonspecific chronic inflammatory expansile lesion.

IPTs can occur in widely varied anatomic locations, being most frequent in the lungs and abdomen. The upper respiratory tract is less commonly involved, with the larynx, trachea, oropharynx, and nasopharynx accounting for only 11% of extrapulmonary cases. The remaining sites in the head and neck account for less than 5% of cases, with involvement of the orbit, paranasal sinuses, major salivary glands, thyroid, and soft tissues of the face and neck occurring in descending order of frequency.
The local symptoms in the head and neck depend on the site of involvement. In extraorbital head and neck locations, the most frequent local symptoms are pain and obstruction of the involved tract. Unlike their counterparts in the visceral organs, sinonasal IPTs cause no systemic symptoms, such as fever, anorexia, weight loss or malaise. The initial symptomatology is a nonspecific sinonasal mass, which progressively worsens over a period of several months or years. The clinical and endoscopic findings may demonstrate a tumefaction covered with normal mucosa, a polyp on the middle meatus, hypertrophy of the inferior turbinate, or hemorrhagic rhinorrhea. These tumors have no sex or age predilection. Other characteristics include bony erosion, remodeling, sclerosis, and thickening.

CT scan of IPT with contrast reveals homogeneous tissue density, with moderate enhancement, which is slightly heterogeneous. On magnetic resonance imaging, IPTs are usually isointense to hypointense relative to muscle on T1-weighted images, with a relatively hypointense T2 signal compared to most other tumors. Contrast enhancement is variable. The radiologic appearance can also be misinterpreted as a malignancy due to bony changes such as erosion, sclerosis and thickening.

A preoperative diagnosis of IPTs is difficult because of the diverse clinical settings in which they arise. Diagnosis of IPT is not possible without histopathologic examination of a tissue sample. The results obtained by fine needle aspiration biopsy or frozen section studies may be unreliable. The unifying histologic feature of this lesion is the highly variable admixture of myofibroblasts and inflammatory cells. The myofibroblasts are characterized immunohistochemically by their expression of vimentin and actin, as well as variable expression of desmin and cytokeratin. The inflammatory component is polymorphous, but lymphocytes, eosinophils, or plasma cells may constitute the major inflammatory component. Mitoses are infrequent. This microscopic description can vary highly, not only between different tumors but even within the same lesion.

IPTs must be distinguished from sinusitis, granulomatous inflammation, collagen vascular diseases, sarcoidosis and neoplastic disorders. In this case, the mass satisfied the histopathologic criteria mentioned above. Staining for amyloid, and cytoplasmic-antineutrophil cytoplasmic antibody (c-ANCA) titer were negative. Multiple myeloma was ruled out by electrophoresis and the negative test for Bence-Jones proteins in urine. All of these possibilities were ruled out in our patient.

Corticosteroid therapy, radiotherapy, chemotherapy and surgery have all been used, either alone or in combination, in the treatment of sinonasal IPT. The treatment of choice for sinonasal IPT is surgery, followed by corticosteroid in cases of incomplete excision. Response to steroid is often unpredictable but these drugs are the primary medical treatment for orbital IPT. The surgical excision must therefore be as complete as possible, yet avoid the principle of radical dissection employed in oncology. Radiotherapy is indicated for patients in whom surgery or corticosteroid therapy is unsuccessful or contraindicated.

Despite the initial clinical and radiologic presentations, sinonasal IPT has a good prognosis. Histologic transformation to an undifferentiated sarcomatous proliferation has not been reported in sinonasal IPT. Unlike orbital localizations, sinonasal IPTs never transform into lymphoma or develop into systemic lymphoma.

Head and neck surgeons must be familiar with the clinical presentations, histopathologic features and management of this rare entity. Its correct recognition by the pathologist is vital to avoid unnecessary treatment such as extensive surgical intervention or radiotherapy.

References

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