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ORIGINAL PAPER

Inflammatory myofibroblastic tumor of the head and neck

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Abstract Inflammatory myofibroblastic tumor (IMT) is a rare benign neoplasm. The aim of this study was to enhance the understanding of head and neck IMT and to improve its diagnosis and management. Clinical features and related treatment of 10 IMT cases were retrospectively analyzed and the literature was reviewed. Tumor sites identified included four in the maxillary space, two in the buccal space, two in the parotid gland, one in the post aurem, and one in the neck. Nine of ten patients received local resection, and one of ten patients received a total maxillectomy. One patient had a local recurrence and died, while the other nine patients had no distant metastases and survived. A computed tomography (CT) exam performed on nine of the ten patients showed that six of these nine cases were heterogeneous in density, while the other three cases were homogeneous. Four cases showed marked heterogeneous enhancement, two cases showed mild heterogeneous enhancement, and three cases showed moderate homogeneous enhancement on contrast-enhanced CT images. The incidence of IMT in the head and neck is low, and local resection is currently the best treatment. A prolonged postoperative follow-up period is necessary for patients with IMT.

Keywords Inflammatory myofibroblastic tumor (IMT) · Computed tomography · Pathology · Prognosis · Survival

Abbreviations
IMT Inflammatory myofibroblastic tumor

Introduction

Inflammatory myofibroblastic tumor (IMT) is a rare benign neoplasm composed of myofibroblastic spindle cells accompanied by inflammatory infiltrate [1]. It is mesenchymal in origin and rarely occurs in the head and neck region. IMT often mimics spindle cell sarcoma, fibrous histiocytoma, or fibrosarcoma in histological appearance. Because of variable cellular composition, IMT is difficult to diagnosis preoperatively through either fine-needle aspiration or biopsy. IMT closely resembles malignant tumors in its biological features, clinical syndromes, and computed tomography (CT) images. IMT underwent histological evolution to a more cellular spindled, round, or polygonal cell pattern. There are few reports studying IMT. Since 2001, ten cases of head and neck IMT were admitted

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to our hospital. Our report of these cases and a review of the literature are aimed at better understanding the diagnosis, treatment, and prognosis of head and neck IMT.

Methods

General data

Ten IMT cases in the head and neck were admitted to the Cancer Center, SUN Yet-Sen University from January 2001 to October 2009. The follow-up period ranged from the date of the definitive diagnosis to the final visit or date of death.

Clinical situation

The painless mass in the parotid gland, neck, post aurem, buccal, and parapharyngeal spaces was the obvious clinical feature of head and neck IMT. Cases showed nasal obstruction, epistaxis, nasal discharge, numbness, face swelling, and headache in maxillary IMT.

CT image

Nine of the ten patients were examined by CT scan. CT imaging was completed with a Twin Flash (Elscint, Haifa, Israel) or a Brilliance TM[16] (Philips Medical Systems, Best, The Netherlands) helical scanner. The scan parameters were 5-mm slice thickness reconstructions, 23-cm field of view, 120 kV, 200 mAs, and 256 × 256 matrix. All nine patients underwent unenhanced and contrast-enhanced CT scans. An intravenous bolus dose of 100 mL of a nonionic iodinated contrast agent (iopromide, Ultravist; Schering) was administered at a rate of 2.5 mL/s for the patients that underwent contrast-enhanced CT scans. Contrast-enhanced CT scans began 50 s following the onset of contrast injection.

Two experienced radiologists reviewed the CT image characteristics of each lesion, which included location, shape, size, number, edge, and attenuation of unenhanced and contrast-enhanced lesions. In the unenhanced CT images, attenuation was classified as hypo-, iso-, or hyper- with respect to the adjacent tissues. In the contrast-enhanced CT images, the degree of enhancement was classified as no, mild, moderate, or marked enhancement.

Results

Clinical data

The clinical data of 10 patients are summarized in Table 1. Six men and four women were included in this study. Age ranged from 9 to 70 years, with a median of 45 years. The most common sites were four in the maxillary space (40%), two in the buccal space (20%), two in the parotid gland (20%), one post aurem (10%), and one in the neck (10%). All of the 10 patients underwent surgical resection without chemotherapy, radiotherapy, or corticosteroids/non-steroidal anti-inflammatory drugs (NSAIDs).

The patients possessed a survival time from 12–90 months with the median being 31 months. One of ten patients died of a tumor recurrence, and the other nine patients are still alive. All ten patients had no distant metastasis.

CT findings

In four cases, the tumors were located in the maxillary sinus; two tumors were on the right and two on the left. In two cases, the tumors were in the buccal space; one tumor was on the right and one on the left. In two cases, the tumor was located in the left parotid gland. One tumor was on the subcutaneous post aurem tissue. The smallest mass in our study was 2.5 × 3.0 cm, and the largest mass was 6.5 × 5.5 cm. The unenhanced CT images showed heterogeneous masses with patchy necrotic foci in six cases, and three small homogenous mass in the remaining three cases. No calcification was observed in any of the cases. The tumors in the maxillary sinus eroded the cavity walls in three cases and the tumor in the deep lobe of the left parotid gland extended into the left parapharyngeal space. The contrast-enhanced CT images showed marked heterogeneous enhancement in four cases, mild heterogeneous enhancement in two cases, and moderate homogeneous enhancement in three cases. All of the masses observed were poorly defined (Fig. 1).

Discussion

Clinical features

IMT is a distinct neoplasm composed of myofibroblastic and inflammatory cells in which mitosis is rare [1]. IMT of the head and neck is rare and most reported cases are isolated incidences from different centers worldwide. IMT is easily misinterpreted as malignant epithelial or mesenchymal spindle cell neoplasms according to clinical and radiological evidence. It is difficult to diagnose because of its complicated pathologic characteristics. IMT was first described by Pettinato when he studied 20 cases of inflammatory pseudotumor [2]. Pathologically, IMT consists of many myofibroblastic cells, fibroblastic cells, and inflammatory cells, which include plasma cells and eosinophils. Due to the multitude of cell types found in IMT, a

Table 1 Summary of clinicopathological features of 10 IMT cases in the head and neck

Case no	Gender	Age (years)	Site	Treatment	Follow-up (months)
1	Female	17	Maxillary sinus	Total maxillectomy	Alive
					No recurrence at 90 m
2	Female	14	Maxillary sinus	Partial maxillectomy	Alive
					No recurrence at 66 m
3	Female	45	Maxillary sinus	Partial maxillectomy	Alive
					No recurrence at 31 m
4	Male	50	Maxillary sinus	Partial maxillectomy	Alive
					No recurrence at 18 m
5	Male	68	Buccal	Buccal tumor extensive resection	Alive
					No recurrence at 20m
6	Female	70	Buccal	Buccal tumor extensive resection	Alive
					No recurrence at 18 m
7	Male	46	Parotid gland	Superficial parotidectomy	Alive
					No recurrence at 31 m
8	Male	9	Parotid gland	Superficial parotidectomy	Alive
					No recurrence at 12 m
9	Male	13	Post aurem area	Post aurem tumor extensive excision	Deadly
					Recurrence four times at 71 m
10	Male	39	Neck (posterior triangle region)	Neck tumor resection	Alive
					No recurrence at 27 m

variety of names were used to describe this disease in the literature, such as inflammatory pseudotumor, plasma cell granuloma, and inflammatory fibrosarcoma.

In 1994, the WHO defined IMT as an intermediate soft tissue tumor that is composed of myofibroblast-differentiated spindle cells and is accompanied by numerous inflammatory cells, plasma cells, and/or lymphocytes [3].

Although IMT occurs at multiple anatomical locations, lesions in the head and neck area are extremely rare. According to our statistics, 33 IMT patients were admitted into our hospital from 2001 to 2008, with only ten patients presenting IMT in the head and neck. IMT in the head and neck shows no specific clinical behavior. However, the cases that occurred in the maxillary sinuses showed unique symptoms such as facial deformation, numbness, exophthalmos, and diplopia due to the destruction of the cavity walls and extension of the mass into the ethmoidal and orbital cavities [4].

CT features

The image findings of IMT demonstrate homogeneous or heterogeneous soft tissue masses, which often extend into adjacent tissue [5]. In contrast-enhanced images, the tumors often demonstrated moderately or marked enhancement [5]. The tumors located in the maxillary sinus often destroyed the bone wall of the cavity [6]. In our series, the six large masses demonstrated heterogeneous density in unenhanced CT images and marked heterogeneous enhancement in four cases and mild heterogeneous enhancement in two cases contrast-enhanced CT images. In contrast, three small masses demonstrated homogeneous density in the unenhanced CT images and moderate homogeneous enhancement in the contrast-enhanced CT images. Three masses in the maxillary sinus destroyed the surrounding bone walls. In the case located in the deep lobe of the parotid gland, the tumor extended into the left parapharyngeal space and demonstrated marked heterogeneous enhancement, similar to findings of malignancy. In view of the above-described image findings, IMT was often misdiagnosed as a malignant tumor.

Therapeutic regimen and prognosis

The mainstay of IMT treatment modality is complete surgical excision [7]. Almost all of the patients of neck, buccal, and larynx IMTs received local excision and recovered successfully. Al-Sindi et al. [8, 9] reported the complete excision of a paranasal sinus mass by a CO_2 LASER while maintaining the maxillary shape as a new treatment modality; the patient manifested no evidence of disease recurrence at a 10-month follow-up visit. In our study, all ten patients underwent excision. Nine patients received local resection, and only one patient with IMT of

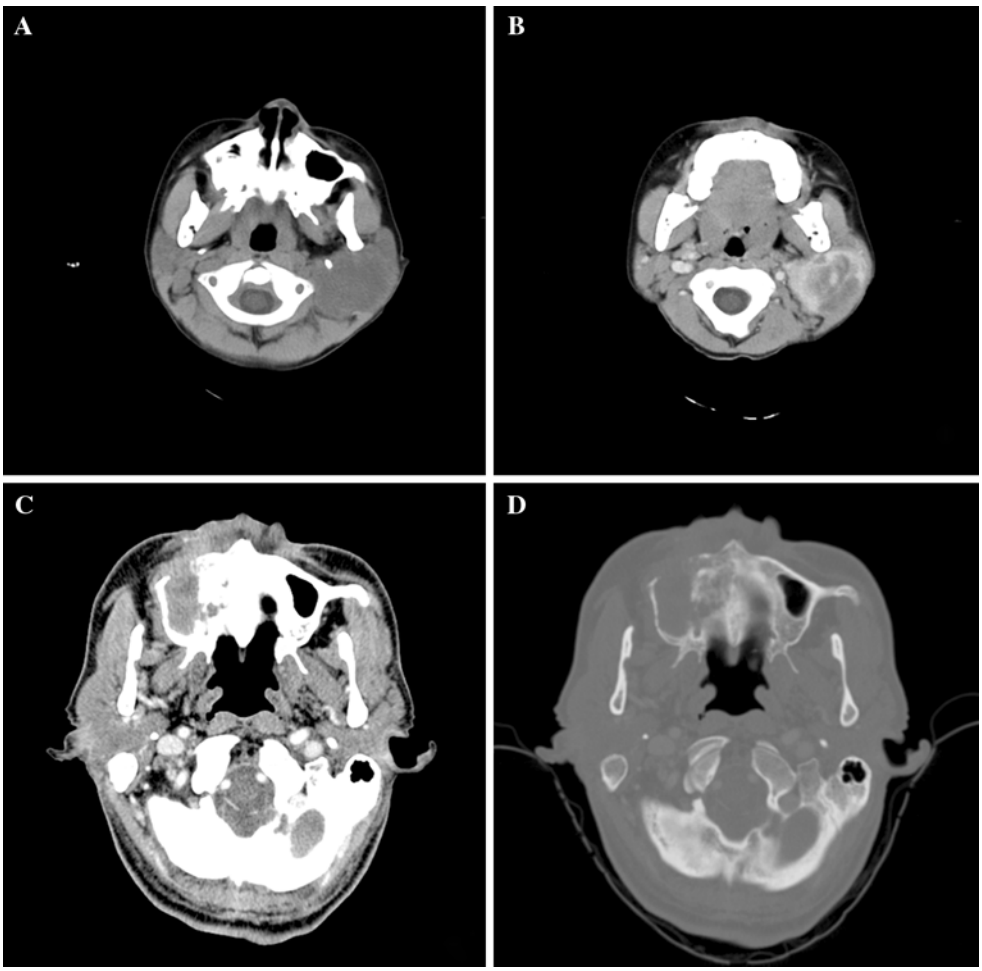

Fig. 1 CT images of IMTs. **a** Heterogeneous density of the tumor in the deep lobe of the parotid gland extended into the left parapharyngeal space in unenhanced CT scan. **b** The tumor demonstrated marked heterogeneous enhancement in contrast-enhanced CT scan. **c** Soft tissue window showed an aggressive mild heterogeneously enhanced mass at the right maxillary sinus. **d** Bone window showed bone destruction of walls of the sinus

the maxillary sinuses received total maxillectomy. One patient had a recurrence of IMT, while no evidence of recurrence was observed in the other nine patients.

IMTs tend to be less aggressive, but have the potential for local invasion and recurrence [9]. In our study, IMT recurred four times in a single patient. Specifically, sinonasal IMT may exhibit aggressive behavior and erode surrounding soft and bony tissues [10]. Thus, sinonasal IMT recurs easily and maxillectomy was performed on the recurrent tumor.

Some medical specialists suggest that high-dose steroids and radiation are important treatment options for IMT. Robert et al. [11] reported that the best approach to managing laryngeal IMT was local excision followed by a high-dose steroid for 6–12 weeks. Thomas et al. [12] reported that a patient with maxillary sinus IMT was treated successfully with a combination of surgery and steroid therapy. Suh et al. [13] suggest steroid therapy as a good choice for IMT treatment. Combined treatment with surgery and corticosteroids is recommended for younger children, especially infants [14]. So, further study is needed to explore the effective nonsurgical treatment modality for IMT.

Conclusion

Currently, the incidence of IMT in the head and neck is low, and local resection is the best treatment modality. IMTs have the potential for local invasion and recurrence. A prolonged follow-up period is necessary after surgical excision.

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Conflict of interests The authors declare that they have no competing interests.

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