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# Clinicopathological analysis of 18 cases of inflammatory myofibroblastic tumor in oral and maxillofacial region

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## Abstract

**Objective** To analyze the clinicopathological features of inflammatory myofibroblastic tumor retrospectively and help maxillofacial surgeons to improve their recognition of its early diagnosis and proper treatment.

**Methods** Data of 18 patients diagnosed with IMT in Nanjing Stomatological Hospital from November 2003 to July 2024 were collected. Their clinical, pathological, imaging features, treatment, and prognosis were analyzed.

**Results** Main clinical manifestations were local masses, 8 cases were accompanied with malignant signs such as pain. No obvious systemic symptoms were reported. Bone destructions were seen in 3 cases. Pathological examination showed that 12 cases were Type II IMT and 6 cases were Type I IMT. 15 cases underwent surgical resections and were followed up for at least 1 years without recurrence.

**Conclusions** The clinical symptoms and imaging manifestations of head and neck IMT are not specific. It is necessary to diagnose IMT by biopsy before operation. **Wide local excision** is the most reliable treatment.

**Keywords** Inflammatory myofibroblastic tumor, Anaplastic lymphoma kinase, Oral and maxillofacial region, Clinicopathological analysis

Inflammatory myofibroblastic tumor (IMT) is a rare tumor consisting of myofibroblastic spindle cells infiltrated by plasma cells, lymphocytes, or eosinophils, along with a mucus-like stroma. It can manifest in various parts of the body, most commonly in the lungs and peritoneum [1]. First identified in the lungs by Brunn in 1939, it was initially termed as an inflammatory pseudotumor by Umiker & Iversin in 1954 [2]. However, due to its histopathological diversity, unknown etiology and pathogenesis, the nomenclature of IMT was confusing. It wasn't until spindle myofibroblasts were recognized as a distinct cell type that the World Health Organization (WHO) officially coined the term "inflammatory myofibroblastoma," defining it as "a tumor characterized by the presence of myofibroblastic spindle cells with a predominant soft tis-

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sue pattern and multiple inflammatory cell infiltrates” [1, 3].

IMT less frequently occurs in the head and neck region, typically presenting as a mass accompanied by pain and other signs suggestive of malignancy. Imaging findings also lack specificity, ranging from a simple soft tissue shadow to accompanying bone destruction. Thus, the nonspecific nature of its manifestations poses significant challenges for clinical diagnosis and treatment. This study retrospectively analyzed clinical characteristics, treatment modalities, and prognosis data from 18 IMT patients to enhance clinicians’ understanding of the disease and facilitate improved diagnosis and treatment strategies.

## Materials and methods

### Patient data

From November 2003 to July 2024, data were collected from medical records of 18 patients diagnosed with IMT at our hospital. Information including age, gender, clinical symptoms, tumor size, surgical method, and other relevant details was compiled for analysis.

### Image analysis

Before surgery, all patients underwent routine CT examinations. Experienced oral and maxillofacial imaging specialists in our hospital provided reports assessing the tumor’s location, morphology, and margins.

### Pathologic analysis

Tumor tissues excised during surgery were fixed in formalin for 24 h, paraffin-embedded, and subjected to hematoxylin-eosin staining and immunohistochemistry. Pathologists issued a joint diagnosis based on these findings.



**Fig. 1** Intraoral Soft tissue mass was presented

## Results

### Clinical data

Among the 18 patients, 11 were male and 7 were female, with a mean age of 42.3 years (range, 9–73 years). 15 cases were located in the soft tissues of the maxillofacial region, while 3 cases involved bony tissues. Clinical presentations primarily included masses or bulges (see Fig. 1), often accompanied by signs suggestive of malignancy such as pain and numbness in 9 cases. None of the patients exhibited systemic symptoms like fever or anemia, and laboratory tests revealed no significant abnormalities. A history of smoking and alcohol use was noted in only one patient (case 18). Details are provided in Table 1.

### Imaging findings

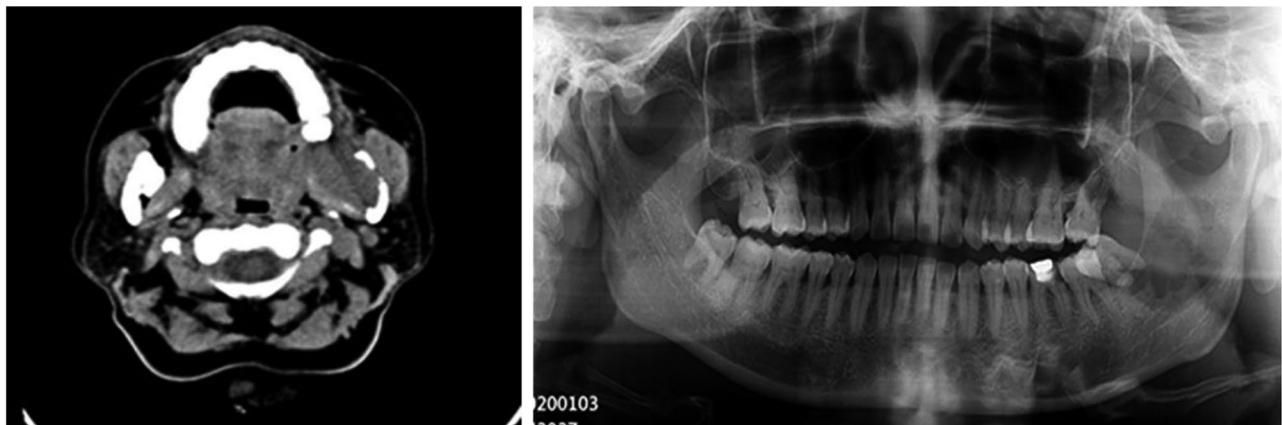
Typically, the lesion appeared as a soft tissue nodule with heterogeneous density. When the mass reached a significant size, it could exert pressure on adjacent bone, potentially resulting in bone resorption or destruction (see Fig. 2). The CT attenuation values typically ranged between 16 and 93 Hounsfield Units (HU). Because the mass was irregular or nodular in shape, its density distribution was inhomogeneous. If the soft tissue mass extended into the muscle, it could cause diffuse enlargement of the affected muscle. Imaging finding of case 5 was partial bone defect on the medial aspect of the right mandibular ramus and inhomogeneous density of the medial pterygoid muscle while no mass was found (see Fig. 3). Case 15 underwent an excisional biopsy of the buccal mass at another hospital, therefore, no significant mass was detected on CT imaging. The summary of imaging results is presented in Table 2.

### Treatment and prognosis

Among the 18 patients, 15 underwent wide local excision of the lesion. In cases where the lesion involved the bone, partial resection or segmental osteotomy of the bone was performed, with subsequent reconstruction of the defect using soft tissue flaps or bone grafts. 2 patients underwent additional wide excision following a confirmed diagnosis by excisional biopsy. 1 patient declined further treatment after diagnosis due to personal reasons. All patients were followed up for a minimum of 1 year. One patient (case 5) initially presented at our hospital in 2015, where a biopsy confirmed the diagnosis of IMT. After discharge, the patient underwent lesion resection at another hospital in 2017. The lesion recurred in 2021, prompting wide local excision and partial jaw resection at our hospital. No recurrence has been observed during follow-up. One patient (case 10) experienced a recurrence 3 years after the initial resection and subsequently underwent wide local excision. Case 3 developed on right oropharynx after IMT excision of left oropharynx 10 years

**Table 1** Basic clinical manifestations of IMT patients

Number	Gender	Age	Occurrence	Location	Symptoms	Size (cm)
1	Male	73	Primary	Right Maxillary Sinus	Gum swelling on the upper right, bleeding on touch	2.5*2*1.5
2	Female	22	Primary	Right Maxillary Gingiva	Bleeding-prone swelling	3*3*1
3	Male	44	Primary	Left Oropharynx	Painful swelling	1*2*1.5
4	Male	50	Primary	Midline of the Floor of Mouth	Painful swelling, enlarged submandibular and submental lymph nodes	4*3*3.5
5	Male	29	Primary	Right Infratemporal Fossa	Pain in the right temporomandibular joint with limited mouth opening	Clinically undetected
6	Female	38	Primary	Left Floor of Mouth	Swelling with left tongue numbness, deviation on protrusion	0.4*1.5*2
7	Female	9	Primary	Right Parotid Gland	Painful swelling	3.5*3*1.5
8	Male	57	Primary	Right Submandibular Gland	Swelling	1.6*0.6*0.7
9	Female	51	Primary	Left Mandible	Swelling, ulceration, pain	3*4*2.5
10	Male	21	Primary	Left Cheek	Swelling, itching	2*2*1.5
11	Female	56	Primary	Left Tongue	Swelling, pain, ulceration	1.2*1.2*1.5
12	Male	41	Primary	Right Mandible	Swelling, pain	7.0*1.5**3
13	Male	15	Primary	Left Buccal Mucosa at Occlusal Line	Swelling	1.5*1.5*1.5
14	Male	56	Primary	Right Cheek	Swelling, poorly defined margins, infiltrative base	2.5*2.5*2
15	Female	45	Primary	Left Cheek	Swelling, pain	2*2*0.5
16	Female	51	Primary	Left Neck	Swelling	3*3*4
17	Male	50	Primary	Right Neck	Swelling	5.6*2.3*4.6
18	Male	54	Primary	Right Tongue	Swelling	1*1*1

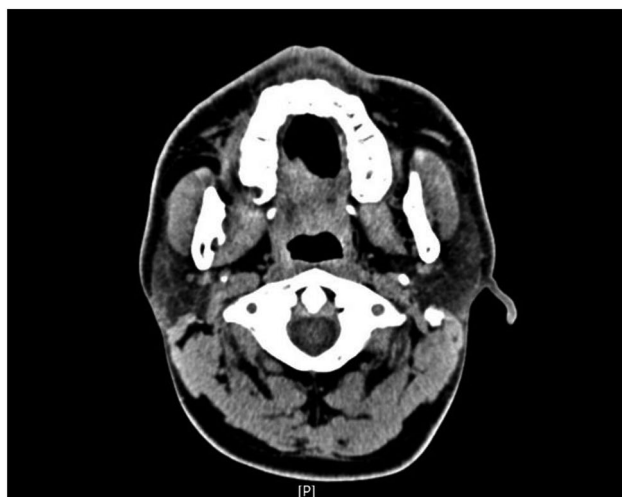
**Fig. 2** Bone destruction was seen in the left mandibular ramus

ago, case 6 developed on the right side of the floor of mouth after excision of IMT on the left side 5 years ago.

#### Pathological characteristics

Microscopically, IMT primarily consists of spindle-shaped myofibroblasts embedded within a mucinous vascular stroma. This is often accompanied by a chronic inflammatory cell infiltrate, predominantly composed of plasma cells and lymphocytes. Among the 16 specimens examined in this study, 12 were classified as type II according to the WHO classification, while 6 were classified as type I. The majority of patients presented with type II IMT.

Histopathological examination revealed that excised masses were typically polypoid or nodular, displaying a soft profile and exhibiting a grayish-white or grayish-brown coloration. The maximum diameter of these masses ranged from 0.4 to 5.5 cm. Microscopic findings from Hematoxylin and Eosin (HE) staining revealed spindle-shaped myofibroblasts distributed unevenly in sheets, bundles, or clusters, with no evidence of nuclear anisotropy or nuclear pleomorphism. Additionally, infiltration of lymphocytes, neutrophils, plasma cells, and histiocytes was observed within the mucinous stroma. Immunohistochemical staining yielded positive results for CD10, bcl-2, and SMA (see Figs. 4 & 5).



**Fig. 3** Partial bone defect on the medial aspect of the right mandibular ramus, and inhomogeneous density of the medial pterygoid muscle

## Discussion

### Etiology

The etiology and pathogenesis of IMT remain elusive. Initially, it was hypothesized to stem from trauma, inflammation, or autoimmune disorders [4]. Viral infections, including herpesvirus and immunodeficiency virus, were also considered potential factors [5], contributing to the

ongoing confusion surrounding IMT's classification. However, with advancements in molecular techniques, approximately 50% of patients have been found to exhibit fusion or rearrangement of the anaplastic lymphoma kinase (ALK) gene with other genes on chromosome 2 at the p23 locus [6, 7]. Notably, the most common fusion involves TPM3, irrespective of IMT location in the head and neck [8, 9]. This discovery strongly supports the notion that IMT is a neoplastic disease driven by genetic alterations that lead to aberrant cell proliferation, rather than solely an inflammatory condition.

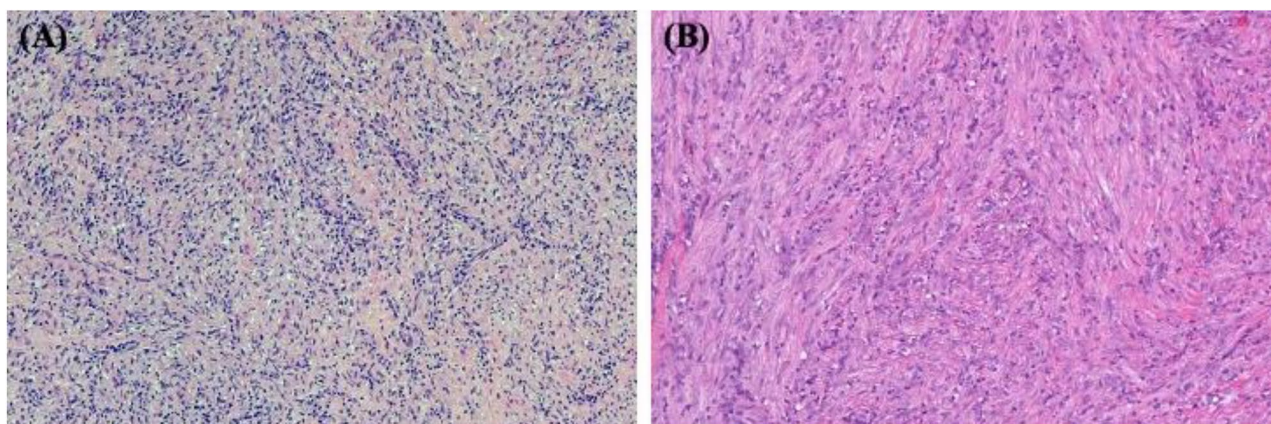
Furthermore, epithelial inflammatory myofibroblast sarcoma (EIMS) represents a malignant variant of IMT characterized by a high incidence of local recurrence and mortality [10].

As a mesenchymal cell tumor with intermediate biological potential, the precise pathogenesis and mechanisms underlying IMT remain obscure. Upon analysis of the patients' medical histories, no significant predisposing factors such as surgery, trauma, medication, or infection, apart from 2 cases of recurrence, were identified. Additionally, patients with a history of trauma or surgery in this study were temporally and spatially distant from the occurrence of IMT. One of the patients had a history of smoking, alcohol abuse, while none of the patients had herpes virus infection.

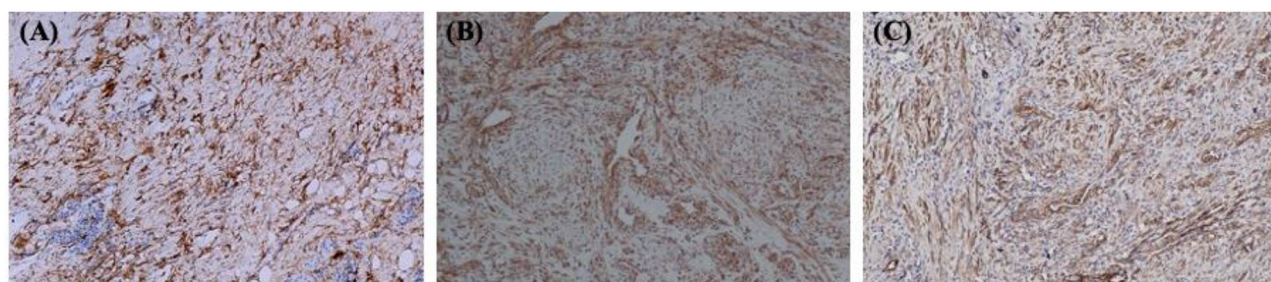
**Table 2** Imaging findings of IMT patients

Number	Location	Imaging findings	CT value (HU)	Maximum cross-section measurement(cm)
1	Right Maxillary Sinus	Inhomogeneous soft tissue shadow; bone destruction	39	4.15*4.02
2	Right Maxillary Gingiva	Gingival mass with bone destruction	33–60	3.15*3.0
3	Left Oropharynx	Soft tissue nodular shadow; Slight deformation and resorption of the alveolar bone	27–71	4.4*3.0
4	Midline of the Floor of Mouth	irregular soft tissue shadow with heterogeneous density and punctate high-density shadows	22–93	4.12*3.1
5	Right Infratemporal Fossa	Partial bone defect on the medial aspect of the right mandibular ramus; inhomogeneous density of the medial pterygoid muscle	Not described	Not described
6	Left Floor of Mouth	Nodular shadow; mildly inhomogeneous density	29–80	2.4*0.8
7	Right Parotid Gland	Soft tissue nodular shadow; compression of the mandibular ramus	29–80	4.15*2.91
8	Right Submandibular Gland	Nodular soft tissue mass with slightly irregular margin	53–72	1.45*0.76
9	Left Mandible	Irregular bone destruction with poorly demarcated boundaries	24–38	4.2*2.9
10	Left Cheek	Roundish soft tissue mass shadow with well-defined borders and heterogeneous density	18–43	1.4*1.3
11	Left Tongue	Slightly thickened soft tissue; irregular surface	48–78	1.45*0.73
12	Right Mandible	Cavitary bone resorption	40	0.67*0.15
13	Left Buccal Mucosa at Occlusal Line	Soft tissue nodular shadow	Not described	Not described
14	Right Cheek	Soft tissue nodular shadow	94	1.41*0.74
15	Left Cheek	No obvious soft tissue mass shadow	Not described	Not described
16	Left Neck	Irregular soft tissue mass with bone destruction	40	3.15*3.1
17	Right Neck	Soft tissue mass with diffuse muscle enlargement	38–56	5.56*4.43
18	Right Tongue	Soft tissue nodular shadow	16–39	0.86*0.8





**Fig. 4** HE staining 20x imaging (A) and HE staining 50x imaging (B)



**Fig. 5** Immunohistochemistry examinations showed positivity for CD10 (A), bcl-2 (B) and SMA (C)

### Clinical manifestations

The clinical manifestations of IMT often lack specificity and are influenced by the site of the mass's occurrence and its characteristics. IMT in the head and neck region often presents with localized symptoms rather than systemic ones. In this study, the majority of clinical manifestations presented as a mass accompanied by pain. If the lesion occurred in the gingiva, it was prone to bleeding. When located in the pterygomandibular space, it could lead to limited mouth opening. If the lesion involved the floor of the mouth, it might cause numbness of the tongue and deviation upon protrusion.

Specifically, IMT in the nasal cavity may cause nasal congestion, bleeding, and loss of smell; in the tonsils and tongue, it may result in difficulty swallowing; and in the orbit, symptoms like tearing and protrusion of the eyeballs may occur [11].

Peng et al. [12] demonstrated that patients with head and neck IMT located in the maxillary sinus, with a tumor size exceeding 4.4 cm, and a preoperative neutrophil-to-lymphocyte ratio exceeding 1.958, were more susceptible to malignant transformation.

### Imaging manifestations

On imaging, IMT may exhibit localized malignant features, although the imaging characteristics of IMT located in soft tissues and bones have also been reported

to resemble those of benign tumors [13]. Typically, CT examinations reveal an expansive, irregular soft tissue mass, often accompanied by dissolution and destruction of surrounding bone. In severe cases, the alveolar bone may disappear, leaving only the basal bone visible [14].

Due to patient cost constraints and limitations in healthcare reimbursement, the cases reported in this study did not undergo MRI examination, which represents a limitation of our work. Some scholars argue that the imaging features of IMTs of the head and neck are non-specific. An ill-defined, aggressive mass and variable enhancement on CT and MRI may suggest the diagnosis of IMT [15], while others maintain that MRI can improve the accuracy of IMT diagnoses and provide critical information for surgical planning [16]. Characteristically, MRI examinations typically depict a solid, irregular mass with low signal intensity on T1-weighted images and enhancement on T2-weighted images. Moving forward, we plan to incorporate MRI into the diagnostic workup of similar cases to enhance preoperative diagnostic accuracy.

Notably, Makoto et al. [17] reported a case resembling a periapical abscess with spontaneous pain and cold sensitivity. Despite initial treatment attempts with root canal therapy and abscess excision, a buccal gingival bulge prompted further biopsy, leading to IMT diagnosis. Similarly, Wang Can et al. [18] demonstrated that IMT in the head and neck region often manifests in the maxilla and

maxillary sinus, displaying diverse appearances, irregular morphology, and indistinct borders on imaging. Notably, calcifications are typically absent, and there may be invasion into neural foramina. However, cervical lymph node metastasis and distant metastasis are rare.

In this study, imaging manifestations primarily consisted of soft tissue nodular shadows, when the mass reached a significant size, it could exert pressure on adjacent bone, potentially resulting in bone resorption or destruction resembling those seen in malignant tumors. The CT attenuation values ranged confirmed the mass density distribution was inhomogeneous. If the soft tissue mass extended into the muscle, it could cause diffuse enlargement of the affected muscle.

### Pathologic manifestations and differential diagnosis

The microscopic composition of IMT comprises proliferating myofibroblasts along with infiltrating lymphocytes, plasma cells, and other inflammatory cells. Based on the proportions of these components, the WHO classifies IMTs into three types [1]: Type I, mucovascular, characterized by loosely arranged myofibroblasts amidst a mucus-like matrix, accompanied by plasma cells, lymphocytes, eosinophils, and angiogenesis. Type II, spindle cell type, featuring densely arranged spindle cells with notable inflammatory infiltration, including scattered plasma cells and lymphocytes, within a variegated mucous matrix. Type III, oligoclonal fibrous type, presenting with collagen sheets resembling scar tissue, densely packed spindle cells, and scattered plasma cell and eosinophil infiltration.

Malignant transformation into epithelial inflammatory myofibroblast sarcoma (EIMS) occurs in only a small percentage of IMT cases. Although one patient in this study exhibited malignant potential, he was not diagnosed with EIMS. Observation of cellular pleocytosis, karyorrhexis, and presence of neutrophils, eosinophils, and multinucleated giant cells necessitates consideration of malignant transformation and the addition of appropriate immunohistochemistry markers such as PCNA, EMA, MyoD1, and p53.

The majority of ALK-rearranged IMT cases are non-malignant. ALK cytoplasmic positivity detected by immunohistochemistry is a common characteristic, serving as one of the diagnostic criteria for IMT. Zhang et al. [19] reported ALK expression positivity in 21 IMT patients (51.2%). While ALK expression detection by FISH or NGS is considered the gold standard, its high cost limits its widespread application [20].

Diagnosis of IMT in oral and maxillofacial region requires differentiation from other tumors characterized by inflammatory infiltrates and myofibroblastic proliferation. Key entities to consider include: 1. fibrosarcoma or malignant fibrous Histiocytoma (MFH): these

tumors exhibit more pronounced cellular atypia and lack the inflammatory cell infiltration typically seen in IMT. 2. Inflammatory Pseudotumor: While similar to IMT, inflammatory pseudotumors are generally benign and lack myofibroblastic proliferation and ALK expression. 3. Solitary fibrous tumor (SFT): SFT typically shows positive immunohistochemical staining for STAT6, whereas IMT is often ALK-positive.

### Treatment

Surgical resection remains the preferred treatment for IMT, with complete resection during the initial surgery being crucial for successful outcomes [21]. Based on the findings of this study, wide local excision is recommended for the treatment of IMT. Excision alone does not significantly reduce the risk of recurrence. A review of recurrent cases in this study revealed that all patients initially underwent lesion excision without wide local excision during their first surgery. Therefore, wide local excision is recommended as the preferred treatment approach for IMT. Optimal surgical margins may differ based on the tumor's location. Pathologically confirmed diagnosis is the prerequisite for determining the resection margin. A review of recurrent cases revealed that one case underwent simple enucleation of the buccal mass without wide local excision due to unclear pathological diagnosis, resulting in recurrence after 8 years. One case of IMT occurred in the pterygomandibular space who was initially treated at another hospital underwent partial mandibular square resection, recurring after 4 years. Upon presentation at our institution, segmental osteotomy with a 2-cm safety margin was performed, and no recurrence has been observed during 2-year follow-up.

Based on our experience, for soft tissue-confined lesions without bony erosion, a 0.5 cm margin is recommended. If glandular involvement is present, concomitant gland resection should be performed, as simple enucleation carries high recurrence risk. For bone-infiltrating tumors, a 2 cm margin is recommended. Thus, achieving negative surgical margins during the procedure is strongly advised as a supplementary method to confirm complete resection. Regarding the indication for postoperative radiotherapy, Song et al. [22] concluded that tumors larger than 5 cm accompanied by ALK overexpression may benefit from postoperative radiotherapy, with recommended dosages. However, since the maximum diameter of our patient's mass was approximately 5.56 cm and margins were negative, postoperative radiotherapy was not recommended, no recurrence was observed during follow-up.

ALK gene rearrangements have opened avenues for targeted therapy using ALK inhibitors such as crizotinib and alectinib. In head and neck IMT, Kazunori et

al. [23] reported successful ALK inhibitor therapy, where a recurrent tumor regressed by 63% after treatment with 600 mg/day of alectinib for 2 months without developing drug resistance. Although therapies targeting fusion genes in IMT have shown promising efficacy, their approval for IMT treatment remains limited.

Anna et al. [24] reported a case of recurrent, lethal, ALK-positive oral IMT where treatment with an ALK inhibitor combined with radiation proved ineffective. Kichenaradjou et al. [25] also reported three cases of maxillary IMT treated with chemotherapy, radiotherapy, and steroids, with one fatality and two patients showing no signs of recurrence at 6 and 9 years postoperatively. Therefore, the efficacy of ALK inhibitors may require further validation through additional studies.

Additionally, some scholars [26] have reported successful treatment of IMT of the maxillary sinus using prednisone and radiotherapy, while others [27] have documented regression of IMT of the mandible with prednisone treatment alone. However, from a clinical perspective, this study has several limitations, including a small sample size, lack of MRI examinations, and insufficient long-term follow-up data. As a rare tumor in the head and neck region, we will continue to collect clinical cases and provide updated reports. We intend to implement MRI as a routine preoperative diagnostic modality. Although some cases have been followed for over 10 years, we will maintain close surveillance, particularly for recently diagnosed and treated cases, including recurrent ones. The clinical symptoms and imaging manifestations of IMT often presented as a mass accompanied by pain in the oral and maxillofacial region, with or without malignant signs. Imaging manifestations primarily consisted of soft tissue nodular shadows, may potentially resulting in bone resorption or destruction resembling those seen in malignant tumors. Hence, the preoperative biopsy is essential to establish a clear diagnosis. Wide local excision remains the most reliable treatment option, with the first surgery aiming for thorough resection in suitable patients. However, extensive resection may significantly impact patients' postoperative quality of life.

For patients who are not suitable for surgery or are intolerant to extensive resection, detecting ALK gene rearrangements can guide treatment decisions. Options may include ALK inhibitors or a combination of radiotherapy. Nevertheless, the lack of a standardized treatment protocol remains a challenge in the field.

#### Author contributions

W. L., M. L., X. H.: Project development, Data Collection, Manuscript writing. Z. F. and Z. C.: Data Collection. J. Y.: Data analysis. X. Y.: Project guidance, Manuscript editing.

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#### Data availability

Data is provided within the manuscript.

#### Declarations

##### Research involving human participants

The protocols and procedures were approved by the institutional review board of the Nanjing Stomatological Hospital and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. All participants provided informed consent, and participants under 16 had informed consent obtained from their parents or legal guardians before their inclusion.

##### Consent for publication

Not applicable.

##### Competing interests

The authors declare no competing interests.

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