Recurrent Inflammatory Myofibroblastic Tumor of Sternocleidomastoid Muscle

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ABSTRACT

Introduction

Inflammatory myofibroblastic tumour (IMT) is a rare intermediate grade mesenchymal lesion with low metastatic potential and a high propensity for recurrence. Most commonly affected sites are lungs, abdomen and pelvis. Head and neck IMTs account for 5% of all IMTs with IMT of sternocleidomastoid muscle being exceptionally rare.

Case Report

A 35 year old female presented with a nodular mass lesion over skin in the left infra auricular area. She had a previous history of swelling at the same site with excision done, and histopathological report revealed inflammatory myofibroblastic tumour. Radiological evaluation was suggestive of neoplastic etiology. Intraoperatively mass was seen over the sternocleidomastoid muscle infiltrating overlying skin. Excision of mass was done along with superficial parotidectomy. Histopathological examination revealed ALK positive inflammatory myofibroblastic tumour of sternocleidomastoid infiltrating the superficial parotid gland.

Discussion

Inflammatory myofibroblastic tumours are extremely rare in head and neck region and have a tendency to recur. Complete resection is the treatment of choice and histopathological examination with IHC is confirmatory for diagnosis.

Keywords

Myofibroblastic Tumor; Sternocleidomastoid; Mesenchymal

According to the World Health Organization (WHO), inflammatory myofibroblastic tumour (IMT) is classified as an intermediate, rarely metastasising mesenchymal tumor which is histopathologically characterised by myofibroblastic spindle cell proliferation with mixed infiltration of plasma cells, lymphocytes and eosinophils.[1] IMT can originate in different anatomical sites with lung being the most common. Extrapulmonary IMT are seen more frequently in abdomen and pelvis, with a predilection for younger patients especially in the first and second decades. Head and neck IMT account for 5% of all IMTs and 14% to 18% of all extrapulmonary IMT.[2,3]

The discovery of rearrangement of the anaplastic lymphoma kinase (ALK) gene demonstrated IMT as a type of true neoplasm instead of an inflammatory reaction. Anaplastic lymphoma kinase (ALK) genes rearrangement have been seen in approximately 50% of IMT and has become a widely accepted marker for IMT.[3,4] Inflammatory myofibroblastic tumours of the head and neck region can clinically mimic malignancy.[8] It is hence warranted that a complete resection of the tumour should be done with histopathological examination and IHC staining for a definitive diagnosis.

Case Report

A 35-year-old female presented with a nodular mass in the left infra-auricular region for a duration of eight months. On taking history, it was found that a swelling had been reported previously at the same site which was excised under local anaesthesia and was reported as inflammatory myofibroblastic tumour, ALK 1 and SMA positive. No

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other details of the surgical procedure were available.

On examination, a nodular mass was seen in the left infra auricular region with overlying skin infiltration (Figure 1). It was mobile and non-tender. Scar from previous surgery was visible and was non-tender.

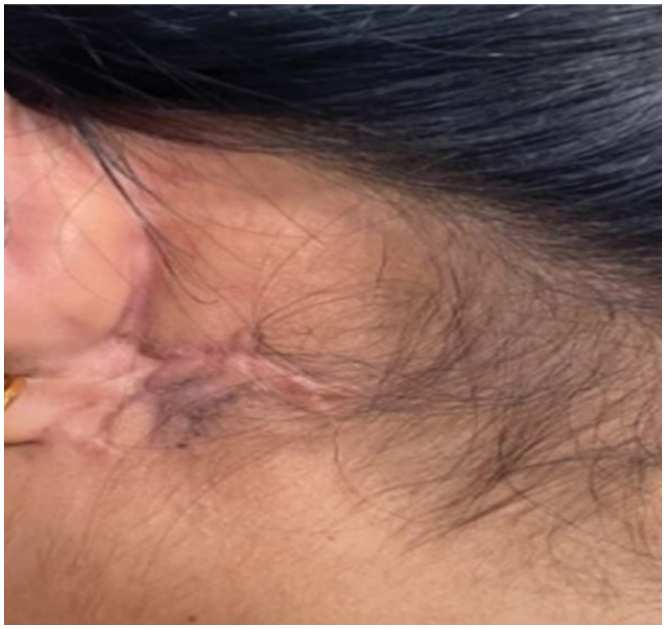

Fig. 1. Clinical Picture Showing nodular lesion

Patient was subjected to a Contrast enhanced computed tomography of the face and neck. The CT scan revealed a heterogeneous enhancing subcutaneous soft tissue density mass lesion measuring approximately 24 x 16 mm in the left parotid region. This lesion had infiltrated into the adjacent superficial lobe of the left parotid gland, the inferior part of the left external ear, and the adjacent sternocleidomastoid muscle. The CT findings were suggestive of an infective or neoplastic etiology.

An MRI of the face and neck revealed a well-defined heterogeneous mass lesion measuring approximately 18 x 13 x 19 mm involving left sternocleidomastoid muscle and skin with infiltration of the adjacent superficial lobe of parotid gland without extension to the deep lobe. The lesion appeared hypointense on T1W, heterogenously hyperintense on T2W, hyperintense on TIRM, and showed minimal post-contrast enhancement. There was suppression on FAT SAT images. (Figure 2)

After pre anaesthetic checkup, the patient was planned for excision of mass under general anaesthesia. A modified Blair incision was given and the SMAS flap was elevated. A firm nodular mass was observed over the anterior

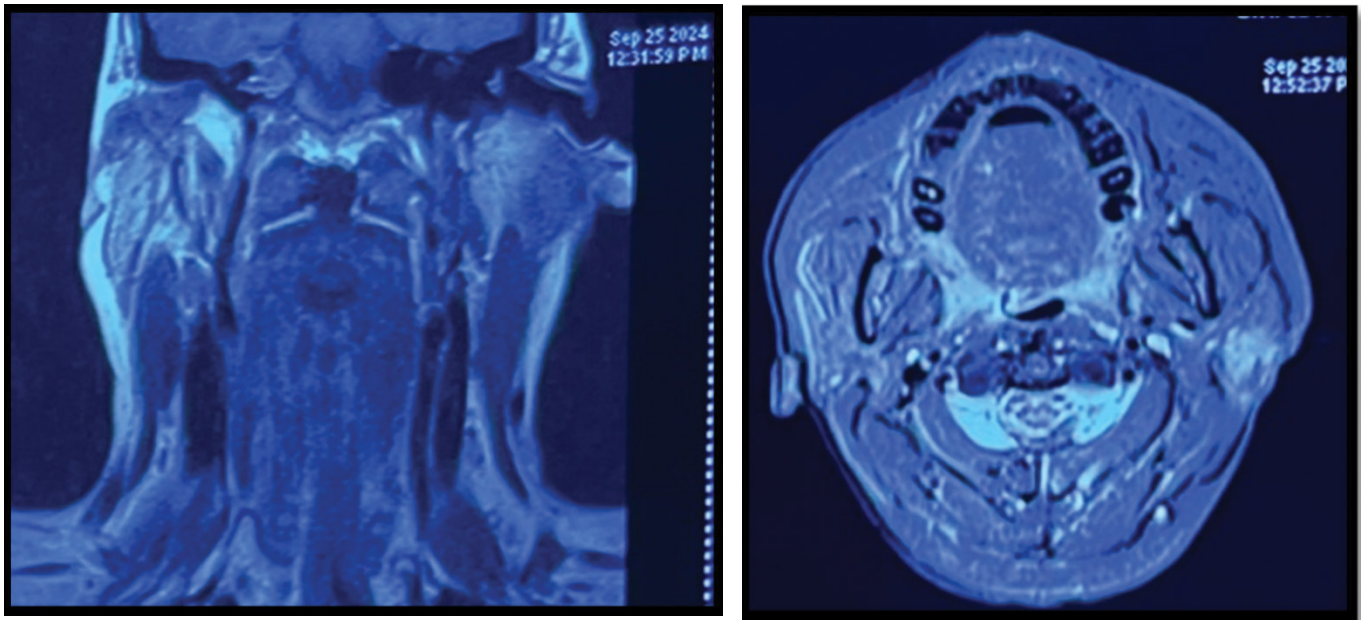

Fig. 2. MRI demonstrating the lesion over left SCM infiltrating skin and adjacent parotid gland

border of the upper one-third of the left sternocleidomastoid along with infiltration of the overlying skin

The lesion was removed in toto with adequate margins and the superficial part of the left parotid gland. Hemostasis was achieved, and the incision was closed in two layers.

Histopathological examination of the surgical specimen suggested an inflammatory myofibroblastic tumour of the left sternocleidomastoid infiltrating into the superficial parotid gland. (Figure 3) Immunohistochemistry revealed ALK positivity (Figure 4)

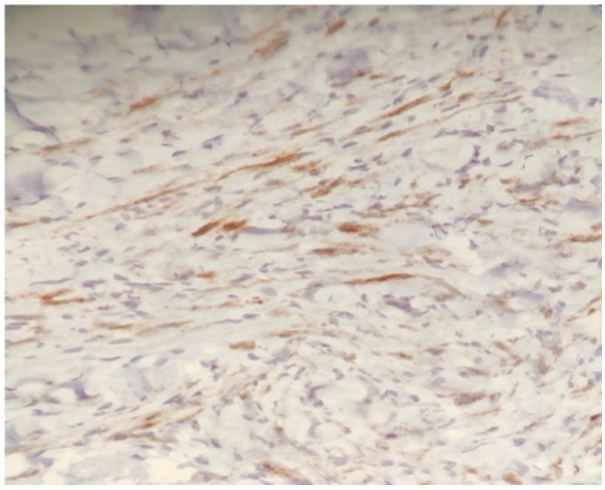

Fig. 3. Granular cytoplasmic ALK positive tumour cells

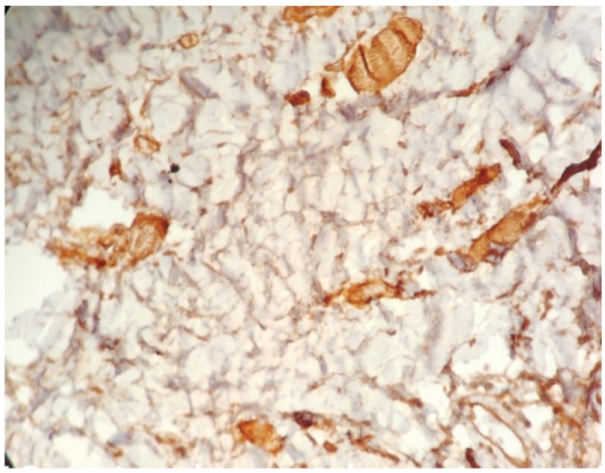

Fig. 4. Desmin positive spindle cells infiltrating skeletal muscle fibres

Discussion

Inflammatory myofibroblastic tumors (IMTs) are rare lesions that present with non-specific clinical features, often making their diagnosis difficult based solely on clinical grounds. While literature suggests that IMTs predominantly occur in children and young adults, typically in the first and second decades of life,[2] our patient presented at the age of 35, which is relatively uncommon.

Imaging is frequently used as initial approach to evaluate head and neck masses, the radiological appearance of IMs in this region can be highly variable. This variability is influenced by the lesion's fibrotic content, degree of inflammatory infiltrate, and stage of disease progression. Consequently, imaging alone is often insufficient for making a definitive diagnosis.[6] Clinico-radiological differentiation of IMT from other malignant tumors can be difficult preoperatively, as observed in our case.

Given these challenges, histopathological examination remains the cornerstone for confirmation. Immuno-histochemistry (IHC) is essential for identifying the myofibroblastic phenotype of these tumors. IMT cells typically express muscle-specific markers such as smooth muscle actin (SMA), desmin (DES), and vimentin. Among these, SMA and ALK (anaplastic lymphoma kinase) positivity are particularly helpful in supporting the diagnosis.[6,7,8] The identification of ALK expression has also opened avenues for targeted therapy using ALK inhibitors.

Emerging research highlights the role of cyclooxygenase-2 (COX-2) in the pathogenesis of head and neck IMT. COX-2–driven inflammation is believed to contribute to tumor invasiveness. Tumors expressing both ALK and COX-2 appear to have a worse prognosis, suggesting a potential benefit from combined ALK and COX-2 inhibitor therapy in adjuvant settings.[4]

Despite advances in molecular therapies, surgical excision remains the primary treatment for head and neck IMTs. The presence of clear surgical margins is the most critical factor in preventing local recurrence.[5] Although IMTs generally have a favorable prognosis, their incomplete encapsulation and proximity to vital structures

in the head and neck can make complete excision difficult, resulting in recurrences. Studies have shown recurrence rates of 31% in ALK-negative and up to 69% in ALK-positive extrapulmonary IMTs. [9] In the present case, initial surgery failed to achieve complete resection, which led to tumor recurrence. A more aggressive excision was subsequently performed. Due to their unpredictable biological behavior, patients with IMT require close follow-up to monitor for recurrence. Our patient is currently disease-free under follow-up.

While several IMT cases have been reported from India, a comprehensive literature review revealed that this is the first reported case of an IMT arising in the sternocleidomastoid muscle with direct extension into the parotid gland. This case underscores the need to consider IMT in the differential diagnosis of atypical neck masses.

In conclusion, extrapulmonary IMTs are rare tumors that require histopathological confirmation and IHC staining for diagnosis. Surgery remains the treatment of choice, although ALK-targeted therapies offer additional options for unresectable, aggressive cases and in adjuvant settings.

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