CASE REPORT

A Large Inflammatory Myofibroblastic Tumor of the Cervical Region Mimicking Malignancy; A Case Report

Ratan Medhi[1], Paramesh Patra[2], Nisha Barala[3]

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ABSTRACT

Introduction: Inflammatory Myofibroblastic(IMT) Tumor is a rare low grade malignant mesenchymal tumor. Most frequently it involves lung and rarely involves head and neck region. The exact etiology is unknown, but recent molecular and genetic evidence suggests it is a true neoplasm rather than purely an inflammatory reaction. Diagnosis is mainly histopathological and surgery is the treatment of choice.

Case report: A 33-year-old female presented to us with asymptomatic large soft tissue mass in the left posterior triangle of the neck for 2 years, which is diagnosed histopathologically as low grade myofibroblastic sarcoma, underwent near total excision, post of HPE report came as inflammatory myofibroblastic tumour, currently patient is on follow up without any recurrence.

Conclusion: Inflammatory myofibroblastic tumor, although a rare soft tissue tumor of head and neck region, but it is locally destructive, and can be confused with other tumors or malignancies because of its nonspecific clinical and radiological presentation. So, clinician should always keep in mind about this rare entity, and if diagnosed correctly then the patient can be cured of the condition just by surgical excision.

Key words: Soft tissue tumour, Neck

INTRODUCTION

Inflammatory Myofibroblastic(IMT) Tumor is a rare soft-tissue tumor[1] histologically characterized by a proliferation of myofibroblast-differentiated spindle cells along with infiltration of various chronic inflammatory cells, such as plasma cells, lymphocytes, and eosinophils.[3] The World Health Organization (WHO) classifies IMT as an intermediate soft tissue tumor or a low-grade malignant, borderline mesenchymal tumor.[7] Although IMT is considered as a benign lesion, it can infiltrate and has destructive behaviors and has a propensity for local recurrence or, infrequently distant metastasis.[5] The exact etiology is unknown, but recent molecular and genetic evidence suggests it is a true neoplasm rather than purely an inflammatory reaction.[3] most common genetic abnormality found in IMT is the rearrangement of the anaplastic lymphoma kinase (ALK) gene on chromosome 2p23, which results in ALK protein overexpression in approximately 50% to 75% of cases.[7] The most frequent location for IMT is the lung.[5] IMTs of the head and neck region are considered rare, accounting for about 5% of total cases.[8] Head and neck IMTs have been reported in various sites including the orbit, paranasal sinuses, larynx, nasopharynx, and the soft tissues of the face and neck.[4] Due to the nonspecific nature of its clinical and radiological features, IMT can often mimic malignant tumors, making pre-operative diagnosis difficult. Therefore, definitive diagnosis relies on histopathological and immunohistochemical examination. Complete surgical resection is the primary treatment modality for IMT.[6]

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Senior Resident[1], Post Graduate Trainee[2,3]
Department of ENT[1-3]
Head and Neck Surgery, AIIMS, Guwahati[1-3]

Corresponding Author: Ratan Medhi
Senior Resident, Department of ENT
Head and Neck Surgery, AIIMS, Guwahati
Email ID: ratan.medhi91@gmail.com

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CASE REPORT

A 33-year-old female presented to us with asymptomatic large soft tissue mass in the left posterior triangle of the neck for 2 years. Initial contrast enhanced MRI shows a well-defined T1 ISO T2 hyperintense lesion measuring approximately 6.9 × 6.9 × 4.9 cm (CC × AP × TR), noted in the posterior aspect of neck which displayed diffuse restriction, a corresponding low ADC value, and homogeneous enhancement on post-contrast study. The mass demonstrated local aggressiveness, involving surrounding musculature, including the left semispinalis capitis, splenius capitis, trapezius, longissimus capitis, and levator scapulae, and was noted to be closely abutting the left common carotid artery and internal jugular vein behind the angle of mandible. CT scan revealed a well-defined non-enhancing soft tissue density mass of size 7.6 × 4.7 × 6.7 cm (AP × TR × CC), noted involving the posterior triangle of left side of neck invading paraspinal muscles and displacing the carotid sheath anteriorly, causing a mass effect over the left internal jugular vein. Trucut biopsy indicated the tissue consisted of interlacing fascicles of spindle cells with mild nuclear atypia and focal lymphocytic aggregates. Immunohistochemistry results from the biopsy showed SMA Patchy Positive staining and a low Ki67 proliferation index of 2%, leading to the initial impression suggestive of low grade myofibroblastic sarcoma. After discussion in the multidisciplinary tumor board patient was plan for neartotal resection of the tumor under general anesthesia. Post op HPE shows fibrocollagenous bundles composed of spindle cells arranged in sheets and bundles, featuring elongated to round nuclei. Focal areas showed dense inflammatory cell collection, characterized by a sprinkling of lymphocytes and plasma cells. There was no evidence of nuclear atypia or abnormal mitotic figures or necrosis found in the sections studied. Immunohistochemistry (IHC) results were supportive of the final diagnosis, with the tumor cells showing strong positivity for Calponin, along with positivity for Beta-catenin, CD 99, SMA (Tram track positivity), and Desmin (Patchy positive). All the features are suggestive of Inflammatory Myofibroblastic Tumour, classified as a spindle cell neoplasm of borderline malignancy. Patient was followed after surgery for last 6 months and wound is healthy without any recurrence or post operative complication.

DISCUSSION

The Inflammatory Myofibroblastic Tumor (IMT) Historically regarded as a reactive process or benign lesion, known by several names including inflammatory pseudotumor, plasma cell granuloma, and inflammatory fibrosarcoma.[1]

The World Health Organization (WHO) currently classifies IMT as an intermediate soft tissue tumor (rarely metastasizing) or a low-grade malignant, borderline mesenchymal tumor, acknowledging its capacity for infiltration, recurrence, and infrequent metastasis.[7]

The precise etiology and pathogenesis of IMT remain largely unknown, although several theories have been proposed, including exaggerated inflammatory responses due to unknown injury, disruptions in immunological responses, or association with infections like Epstein—Barr virus.[8] Modern cytogenetic and molecular findings consider IMT as a true neoplasm rather than purely an inflammatory reaction.[5,7] A hallmark molecular feature in many IMTs is the rearrangement of the anaplastic lymphoma kinase (ALK) gene and abnormal ALK expression promotes tumorigenesis by inducing abnormal proliferation of myofibroblastic cells and activating cell proliferation pathways.[5] In one study focusing on head and neck IMT, cycIooxygenase-2 (COX-2) expression seen in 100% of the cases. IMT is rare overall, constituting less than 1% of all body tumors, with the predominant location being the lung.[2] Head and neck IMT accounts for about 5%

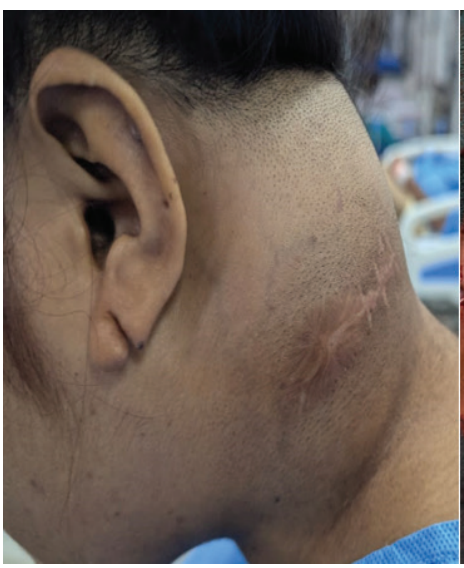

Fig 1: Pre operative image showing the neck swelling

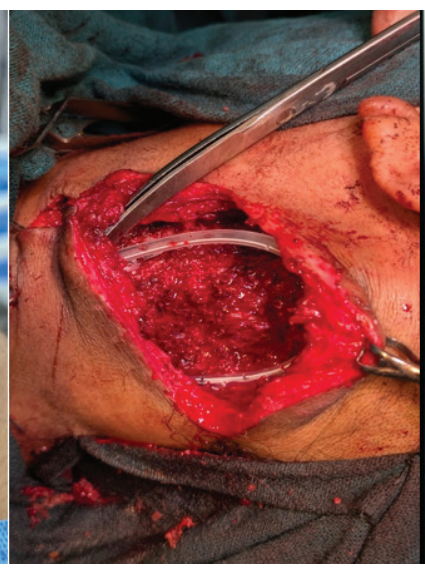

Fig 2: Intra operative image after near total excision

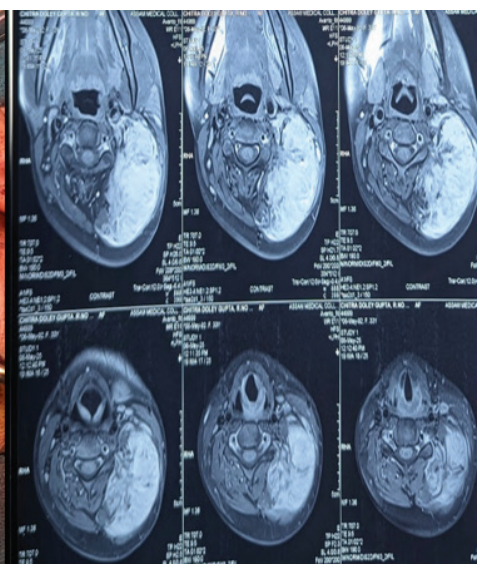

Fig 3: Contrast enhanced MRI neck

of all IMTs. These tumors are overrepresented in children and young adults, although they can affect individuals of any age.[1] Presentation is highly dependent on the location and is often non-specific, frequently manifesting as a mass lesion that may be painful or painless, progressing over months or even years.[8] Pre-operative diagnosis is often difficult because the clinical and radiological manifestations of IMT are nonspecific, frequently mimicking malignant tumors. Imaging study particularly CT and MRI, plays important role in characterizing the tumor. Computed Tomography (CT) IMT is characterized as a soft-tissue density mass, often with variable degrees of enhancement after contrast administration. CT is effective for showing bone changes, which may include erosion, scalloping, or sclerosis, further contributing to the misdiagnosis as a malignant lesion. Magnetic Resonance Imaging (MRI) is generally superior to CT due to its better soft tissue contrast resolution and is recommended for evaluating HNIMTs.[2] Definitive diagnosis requires histopathological examination, which is considered the gold standard.[3] Grossly, the mass is typically nodular, possibly well-defined or non-encapsulated.[7] Microscopically, IMT is defined by the proliferation of spindle cells (myofibroblasts/fibroblasts) set in a collagenized or myxoid stroma, accompanied by a mixed infiltrate of chronic inflammatory cells, notably lymphocytes, plasma cells, and eosinophils.[8] Histologically, three main patterns are recognized: (I) a myxoid-vascular pattern, (II) a compact spindle cell pattern (with cells arranged in fascicles/storiform patterns), and (III) a hypocellular fibrous pattern.[6] IMT tumor cells characteristically express antigens indicating myoid differentiation, being strongly positive for Vimentin. They are also typically positive for Smooth Muscle Actin (SMA), and variably positive for Desmin and Calponin. While ALK positivity is common and helpful, a negative ALK status does not exclude the diagnosis of IMT.[6] In our case IHC positivity seen for Calponin, catenin, CD99, SMA and Desmin. Due to its infiltrative and potentially destructive behavior, complete surgical resection is the gold standard and the primary therapeutic approach.[3,8] However, the local recurrence rate for incompletely resected HNIMT can be as high as 50%.[8] For inoperable tumors, or cases with positive margins, radiotherapy, chemotherapy, or steroids may be utilized.[2] The detection of the ALK fusion protein has opened avenues for targeted therapy; ALK inhibitors like Crizotinib have shown effectiveness in treating ALK- positive IMTs, particularly those that are difficult to resect or have recurred.[8]

Despite its complexity and tendency to mimic malignancy, IMT of the neck generally carries a good prognosis if completely resected, with metastasis being rare (less than 2%).[1] Continued long- term follow-up is necessary due to the risk of local recurrence.

CONCLUSION

Inflammatory myofibroblastic tumor, although a rare soft tissue tumor of head and neck region, but it is locally destructive, and can be confused with other tumors or malignancies because of its nonspecific clinical and radiological presentation. So, clinician should always keep in mind about this rare entity, and if diagnosed correctly then the patient can be cured of the condition just by surgical excision

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