

# Angiomyxolipoma of Right Cheek: A Rare Case Report

RUPALI BAVIKAR<sup>1</sup>, APEKSHA SHARMA<sup>2</sup>, CHARUSHEELA GORE<sup>3</sup>, BHASKAR BHARDWAJ<sup>4</sup>

## ABSTRACT

A lipoma is a benign tumour of adipose tissue and is the most common soft-tissue tumour in adults. It is typically a well-encapsulated, slow-growing, painless mass composed of mature fat cells. Lipomas can develop anywhere in the body, but they are most frequently found in the arms, shoulders, back, neck, and thighs. Angiomyxolipoma (AML) is a very rare variant of lipoma that consists of an admixture of adipose and myxoid elements with numerous vascular structures. Hereby, the authors present a case of a 56-year-old male who presented with a solitary swelling in the right cheek that had been present for the last two years. The Fine Needle Aspiration Cytology (FNAC) findings showed a few bland spindle cells along with some areas of dilated benign cells with elongated nuclei. Histopathological examination revealed a well-encapsulated tumour composed of mature adipose and myxoid tissues, along with blood vessels, confirming the diagnosis of AML of the right cheek. AML is a rare, benign soft-tissue tumour composed of adipose, myxoid, and vascular components. It is considered a variant of lipoma and is typically slow-growing, painless, and well-circumscribed. As of recent literature, fewer than 20 cases of AML have been reported in the English medical literature. Due to their rarity and histological overlap with other myxoid and vascular-rich tumours, such as myxoid liposarcoma or angiomyxoma, accurate diagnosis is crucial. Immunohistochemical staining and careful histopathological evaluation are essential to distinguish AML from other entities.

**Keywords:** Angiomyxoma, Benign, Lipoma, Liposarcoma

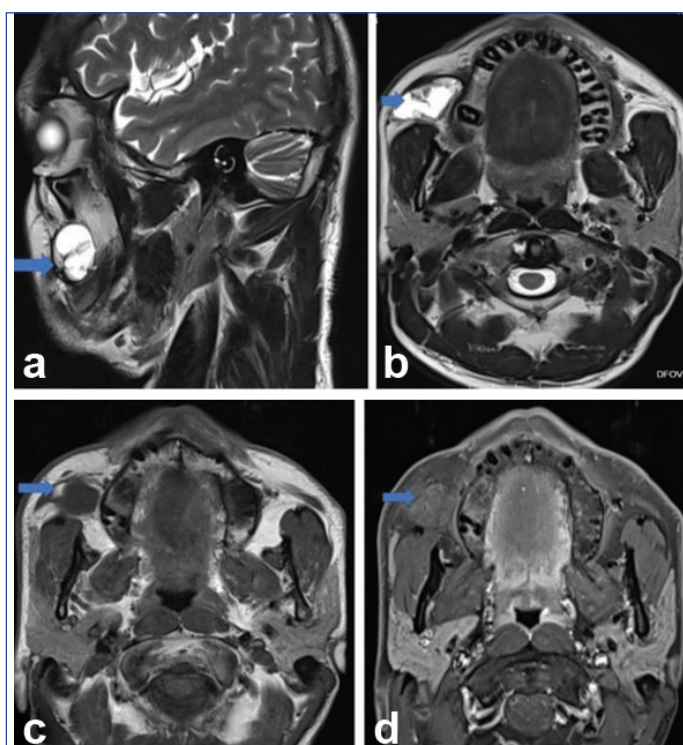
## CASE REPORT

A 56-year-old male presented with complaints of swelling over the right cheek for the past two years. The swelling was gradually progressive in nature and painless. There was a history of tobacco chewing for the last 20 years. The patient had experienced a similar swelling on the right cheek eight years ago, which was excised; however, no documentation is available from the previous excision. The patient reported no significant family history and no other co-morbidities. He had consulted surgeons and dermatologists regarding this swelling, who advised him to undergo excision of the mass.

On examination, there was a single, fixed, non tender swelling on the right upper lateral cheek measuring approximately 2×2 cm. An MRI of the neck, both plain and with contrast, was performed, which included multiplanar multi-echo sequences. The Magnetic Resonance Imaging (MRI) revealed a well-defined T2 hyperintense lesion measuring 24×16×26 mm, located anterior to the right masseter muscle beneath the right zygomaticus major. This lesion was compressed and displaced anteriorly, displaying septae and foci of blooming, with progressive enhancement noted on the contrast study, suggestive of a haemangioma [Table/Fig-1a-d].

The patient underwent ultrasound-guided Fine Needle Aspiration Cytology (FNAC). The microscopic examination of the smears showed red blood cells in the background and a few areas of dilated benign-looking cells with oval to elongated nuclei. No evidence of malignant cells was found in the examined smears. Based on these features, the diagnosis of a benign spindle cell lesion was made. The patient was advised to undergo excisional biopsy for a confirmative diagnosis [Table/Fig-2].

The patient underwent surgical excision under general anesthesia. A wide local excision was performed. Grossly, authors received a right cheek swelling measuring approximately 3.5×2.5×2 cm [Table/Fig-3]. On microscopy, Haematoxylin and Eosin (H&E)-stained sections showed a well-encapsulated tumour composed of mature adipose tissue and myxoid tissue. Both components exhibited the

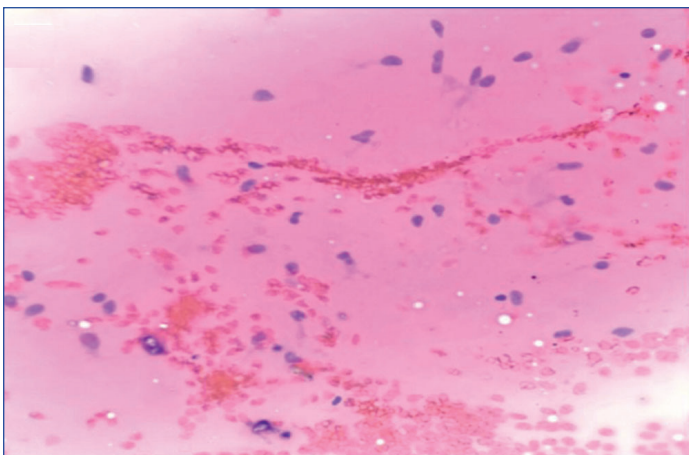


**[Table/Fig-1]:** Photograph showing MRI film of neck (Plain (a,b) and Contrast (c,d)) showing a fairly well defined lesion anterior to right masseter muscle beneath right zygomaticus major suggestive of cavernous haemangioma (blue arrow).

presence of thick and thin blood vessels. There was no evidence of atypia or malignancy [Table/Fig-4]. Therefore, based on these features, it was diagnosed as an AML of the right cheek. The patient has been on regular follow-up post-excision for the last year and currently has no complaints.

## DISCUSSION

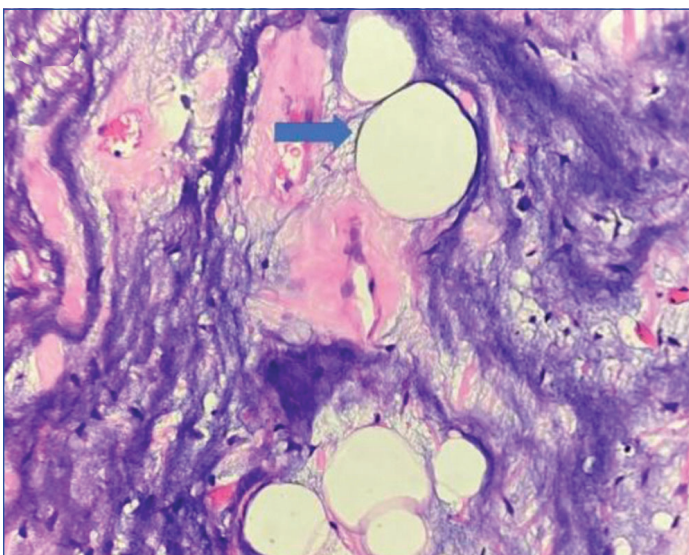
Lipomas are extremely common, but their variants are rare. Different types of lipomas are categorised based on their location and the



**[Table/Fig-2]:** Fine needle aspiration showing moderately cellular smears with Red Blood Cells (RBCs) on the background and few areas of bland spindle cells. (Leishman stain, x200).



**[Table/Fig-3]:** Photomicrograph showing the gross picture of the swelling on the cheek. It has gel like consistency.



**[Table/Fig-4]:** Photomicrograph showing a well encapsulated tumour composed of mature adipose tissues (blue arrow) and myxoid tissue with presence of blood vessels. (H&E stain, x400).

presence of peculiar features in histopathological examinations [1]. AML is a rare variant of benign lipoma with characteristic histopathological and immunohistochemical features. It consists of fatty tissue admixed with myxoid stroma and blood vessels. Confirmatory diagnosis is based on radiological and histopathological correlation. The age group typically affected is middle-aged patients, predominantly male. The location is the subcutaneous tissue, although a few cases have been reported on the spermatic cord and in the subungual region [2]. Only 20 cases have been documented in the English medical literature, with just three involving the spermatic

cord. Due to its rarity, precise incidence rates for AML are not well established [3,4]. Histopathological findings reveal an admixture of a myxoid area with a few mature adipose tissues and numerous thick-walled structures.

The primary differential diagnoses include myxoid liposarcoma, angiomyolipoma, and spindle cell lipoma [5]. Myxoid liposarcoma is an important consideration due to its malignant potential; however, it exhibits increased cellularity, lipoblasts, and characteristic chromosomal translocations (e.g., the FUS-DDIT3 fusion gene), which are absent in Angiolipomas (AML) [6]. Benign spindle cell lesions represent a key differential diagnosis and are a general term for a group of benign tumours composed of spindle-shaped cells (elongated nuclei). Examples include nodular fasciitis, neurofibroma, fibroma, and myofibroblastoma.

On microscopy, many spindle cells are observed, and the stroma is usually fibrous or collagenous, sometimes mildly myxoid. Vascularity is variable and typically not prominent, and adipose tissue is rare. In contrast, AML has a triphasic component: the main cell types are adipocytes, spindle cells, vascular elements, and myxoid matrix, with a prominent myxoid or mucoid stroma [7]. It is highly vascular, with thin-walled or thick-walled vessels and a well-developed mature adipose component. Given its benign nature, the prognosis is excellent, and malignant transformation has not been reported. However, long-term follow-up may be advisable in cases with incomplete excision or uncertain histopathological features [8].

The AMLs share features with myxoid liposarcoma, angiomyolipoma, and other myxoid neoplasms. Immunohistochemistry provides additional diagnostic confirmation through the detection of specific cellular markers [9,10]. The key immunohistochemical markers for AMLs include Vimentin, Cluster of Differentiation (CD)34, S-100 protein, Desmin, Smooth Muscle Actin (SMA), Human Melanoma Black-45 (HMB-45) and Melan-A. Vimentin is strongly positive in AMLs, indicating a mesenchymal origin. CD34 is often positive in vascular and stromal components, helping to differentiate it from other myxoid tumours. S-100 protein is expressed in adipocytic components, assisting in distinguishing AMLs from non lipomatous myxoid tumours. Desmin and SMA are typically negative, ruling out myogenic tumours and angiomyolipomas. HMB-45 and Melan-A are negative, differentiating AMLs from angiomyolipomas, which express these melanocytic markers.

## CONCLUSION(S)

Due to its rarity, AML may be misdiagnosed as other soft tissue neoplasms. Imaging studies, including ultrasound, Computed Tomography (CT), and MRI, can aid in the preoperative diagnosis, but histopathological examination remains the gold standard for definitive identification. Surgical excision is typically curative, with an excellent prognosis and low recurrence rate. Awareness of this entity among clinicians and pathologists is crucial for accurate diagnosis and appropriate management. Histopathological examination plays a vital role in the definitive diagnosis of AML. Given its rare occurrence and histological overlap with other soft tissue tumours, imaging alone may not provide a conclusive diagnosis. Histopathology allows for the identification of the tumour's characteristic components adipose tissue, myxoid stroma, and vascular structures helping to distinguish it from other benign and malignant neoplasms. Additionally, immunohistochemical staining can further aid in differentiation. Accurate histopathological evaluation ensures appropriate treatment planning, prevents misdiagnosis, and helps determine the tumour's benign nature, reducing unnecessary interventions.

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