Infiltrating (Intramuscular) Angiolipoma of the Foot in a Pediatric Filipino Patient: A Rare Case Study Assessing Clinical, Imaging, and Surgical Findings

Abstract

Angiolipomas, characterized by mature adipose tissue intermixed with small vessels, are rare benign tumors that are most commonly found in the extremities and typically manifesting between the second and third decades of life. Here, we report a unique case of Infiltrating (Intramuscular) angiolipoma in a 3-year-old Filipino boy with a foot mass, highlighting the rarity of this presentation among Filipino children and the unusual location in the foot. The discussion delves into the histological and radiological features, the differential diagnosis, and treatment modalities, with surgical excision being the primary approach. In conclusion, recognizing this entity, especially in atypical locations and patient demographics, is crucial for accurate diagnosis and management, adding valuable insights to existing literature. Surgical excision offers favorable outcomes, emphasizing the importance of meticulous technique in achieving complete tumor removal. Histological confirmation is essential, along with long-term follow-up to monitor for recurrence. This case underscores the need for continued vigilance in recognizing and managing rare soft tissue tumors, ensuring optimal patient care and outcomes among our patients.

Introduction

WHO defines Angiolipoma as a subcutaneous tumour consisting of mature fat cells intermingled with samll and thin-walled vessels, a number of which contain fibrin thrombi[1]. Bowen initially documented this phenomenon in 1912. These growths make up a range of 6% to 17% of all lipomas[3]. The limbs, typically the forearm, are the primary location, followed by the trunk, with manifestations typically occurring in the late teens or early twenties[1].

In 1966, J. J. Lin and F. Lin delineated two variations of angiolipomas, categorized as infiltrating and noninfiltrating (encapsulated) types. Their distinction is crucial due to distinct biological behaviors. They outlined criteria for identifying angiolipomas, including: (1) observable tumor formation with or without a capsule, (2) microscopic confirmation of mature lipocytes comprising the majority (at least 50%) of the tumor, and (3) microscopic evidence of angiomatous proliferation within the tumor[3].

There exist two forms of angiolipoma: noninfiltrating and infiltrating. Noninfiltrating angiolipoma typically affects young individuals, presenting as tender, soft nodules on the skin. Occasionally, compression of nerve fibers alongside vascular channels may be observed. Treatment usually involves simple removal. Infiltrating angiolipoma, although rare, has been documented in only 23 cases in English literature. Although histologically benign, this tumor has the capacity to infiltrate neighboring tissues including bone, muscle, nerves, and fibrous tissue, resulting in unusual clinical manifestations that may mimic those of malignant tumors. Complete excision, encompassing normal tissue around the tumor, is essential. Radiotherapy may be necessary in cases of recurrence. The foot represents an uncommon site for angiolipoma, with only five cases reported since 2022[5]. To our knowledge, there has been no reported cases among Filipino children.

Case Report

This case involves a 3-year-old Filipino male who presented with a mass on his right foot. The mass initially appeared two years ago as a 1cm lesion on the dorso-lateral aspect of the midfoot and has since gradually increased in size. Upon consultation, the mass measures approximately 16x13cm, displaying a soft to firm consistency and non-tenderness upon palpation. It is immobile over the dorsum of the right foot, with no associated lymph node enlargement in the popliteal or inguinal regions. Additionally, there is observed plantarflexion of the 2nd-4th toes and limited dorsiflexion of the ankle, yet no sensory deficits are evident, and capillary refill time remains normal. Importantly, there is no discernible impact on function or evidence of neurological abnormalities. Past medical history, family history, neonatal history, and general physical examination findings are unremarkable.

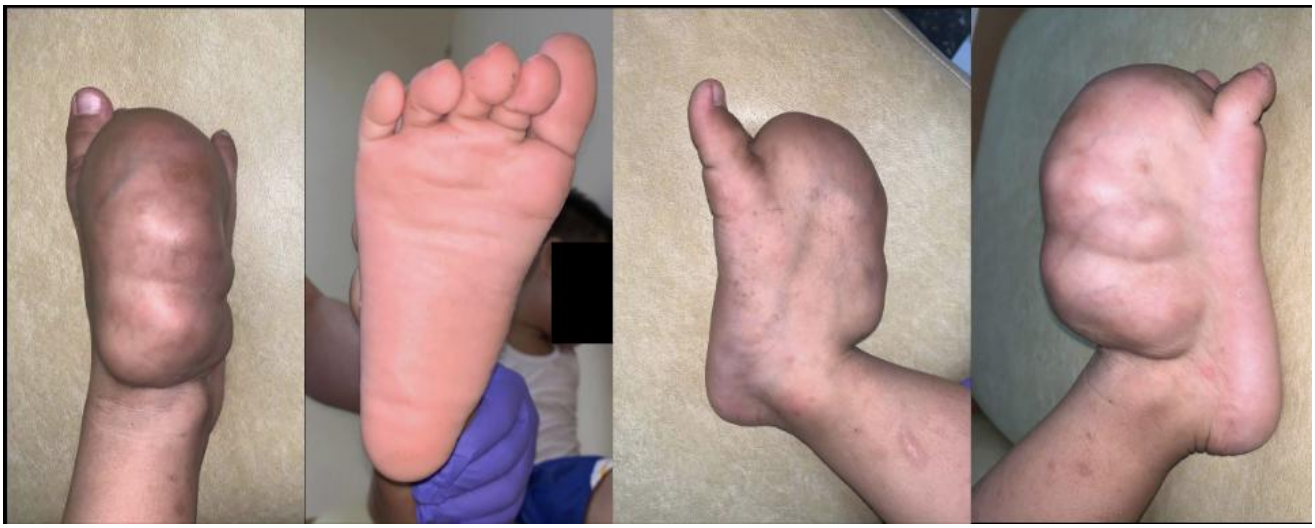

Fig. 1 Gross Imaging of the foot of a 3 year old Filipino boy with a mass located in the dorsum of the foot.

The foot radiograph revealed a sizable, lobulated soft tissue mass density located dorsally on the right foot. There were no notable changes in bone remodeling observed between the right second to fourth metatarsal bones, with the most pronounced effect noted on the second metatarsal bone, characterized by cortical thinning.

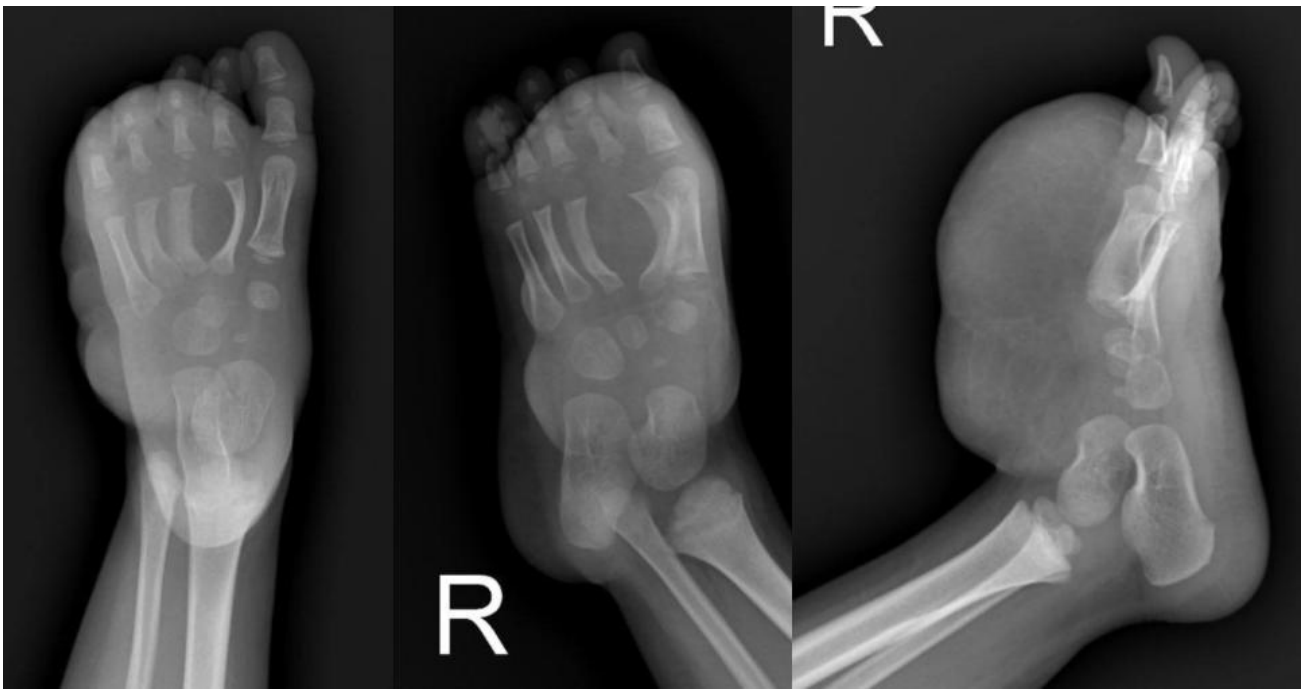

Fig. 2 Radiographic imaging of the Right foot showing a large soft tissue mass on top of the foot with minor thinning of the cortex in the second metatarsal bone.

The MRI findings revealed a large, lobulated structure exhibiting T1-weighted hyperintensity, with a reduction in signal observed on fat suppression sequences. Within this structure, multiple enhancing septations were noted. Erosive changes were observed in the underlying metatarsal bones, yet there was no extension into deeper soft tissues. Joint alignment appeared normal, and marrow signal remained intact. The sesamoid bones showed no remarkable features, and there was no evidence of joint effusion or abnormal fluid collection. Additionally, no muscle atrophy was detected, and both flexor and extensor tendons appeared normal in size and signal. Integrity was maintained in the Lisfranc ligaments and interphalangeal collateral ligaments, with no disruption noted in the plantar plates. Moreover, the plantar fascia did not exhibit thickening.

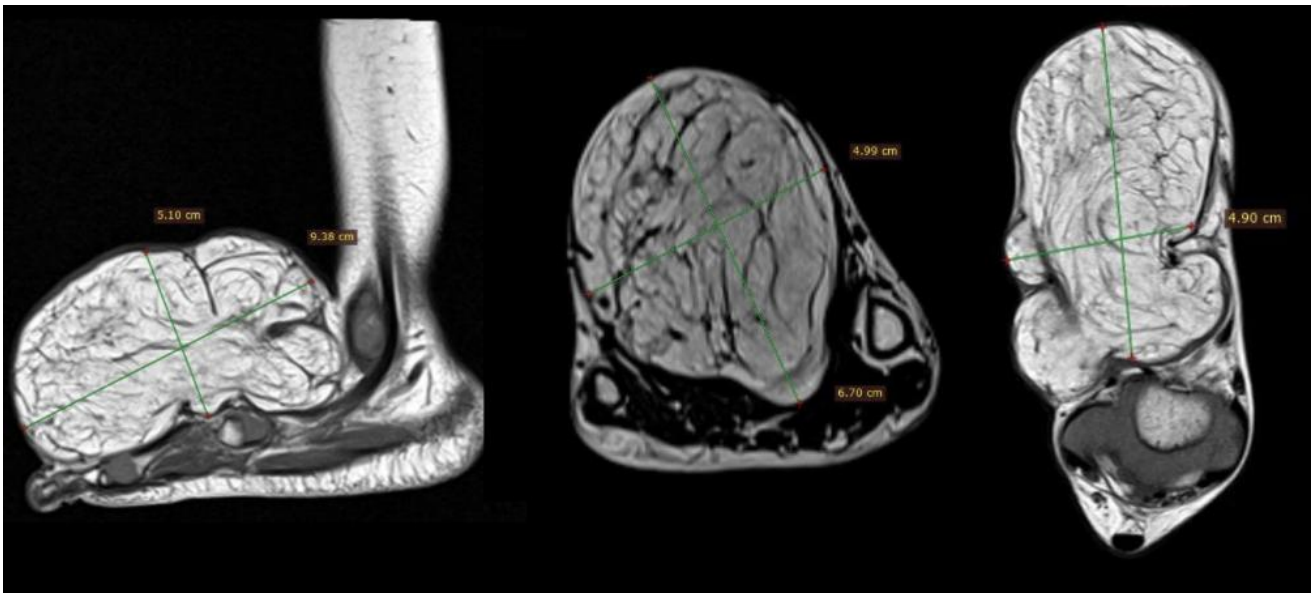

Fig. 3 MRI imaging of the right foot showing bright signals on T1-weighted images, but dark signals when fat was suppressed, with several dividing walls inside; while there were changes to the metatarsal bones underneath, they didn't reach deeper tissues.

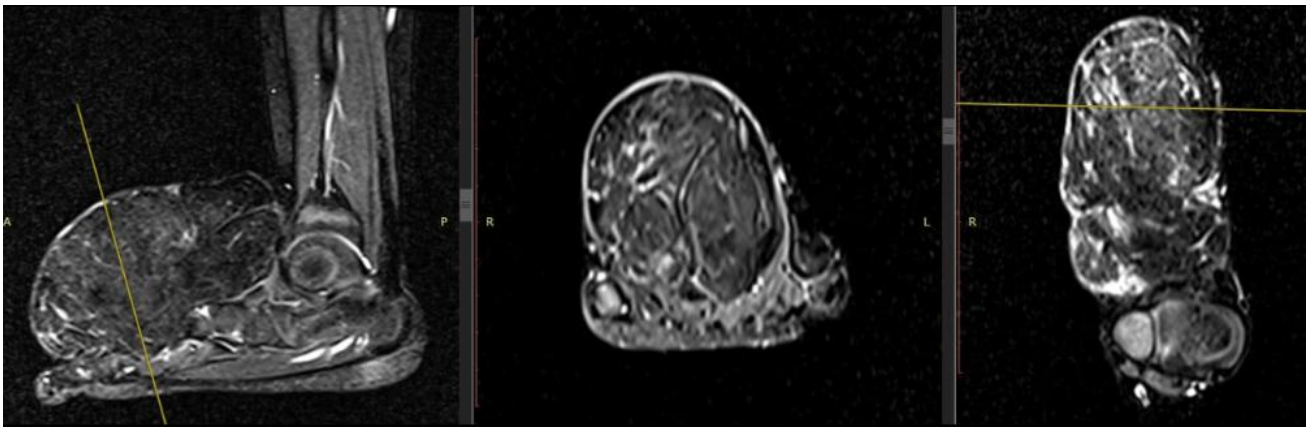

Fig. 4 Joint alignment, bone marrow, sesamoid bones, joint fluid, muscles, and ligaments all appeared normal, with no signs of damage or abnormality on T2 weighted imaging of the right foot.

Based on the clinical presentation, the initial list of potential diagnoses included Lipoma, Liposarcoma, and Rhabdomyosarcoma. After obtaining anesthesia and pediatric clearance, the patient underwent an incisional biopsy of the mass. A 2 cm transverse incision was made at the most distal dorsal mass, revealing a lobulated, yellowish lesion interspersed with dark-red structures suspected to be vessels. Samples were collected and sent for histopathological examination, which ultimately confirmed the presence of Intramuscular Angiolipoma.

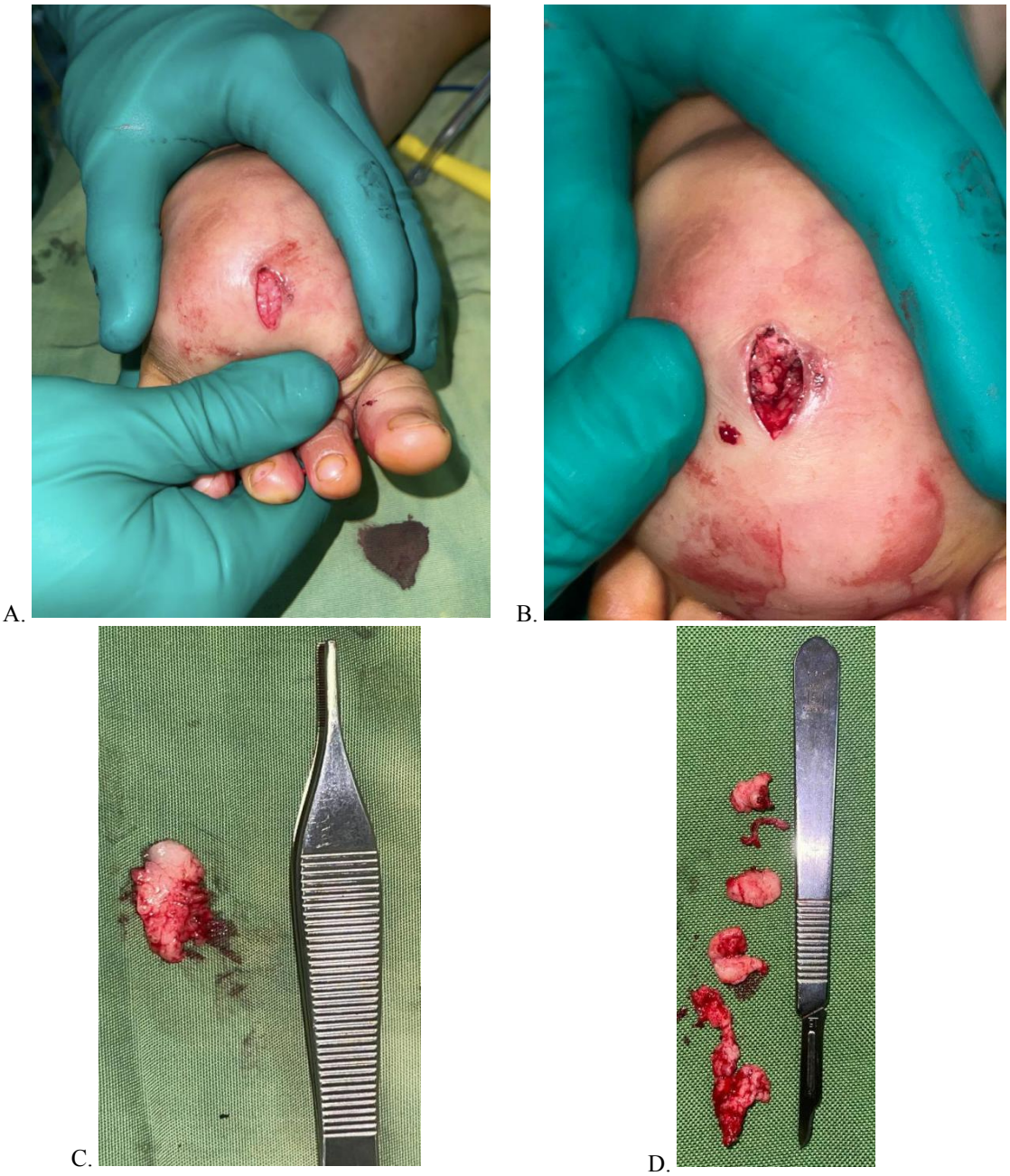

Figure 5: Incisional Biopsy of the Foot Mass. A. A 2 cm longitudinal incision made at the distal dorsal foot mass. B. Deep incision revealing septations within the mass. C. Superficial incision displaying a white to yellow mass with the convergence of vessels. D. Biopsy samples collected for Histopathological testing.

Subsequently, the patient underwent another operation, during which en bloc resection of the mass, along with a normal rim of surrounding tissue, was performed. The mass displayed an ill-defined margin and was not bordered by the deep intermetatarsal ligament. Excision of the mass was meticulously executed while preserving neurovascular structures, followed by layered closure of the wound. The excised mass underwent further histological evaluation, revealing mature adipose tissue intertwined with proliferated vascular elements devoid of atypical features. Thick-walled blood vessels with collagen deposition causing luminal obstruction and occasional capillaries with fibrin thrombi were observed, consistent with the features of angiolipoma. No additional immunohistochemistry analysis or genetic marker studies were conducted. The entire margin was meticulously evaluated, confirming the final diagnosis of Infiltrating (Intramuscular) Angiolipoma.

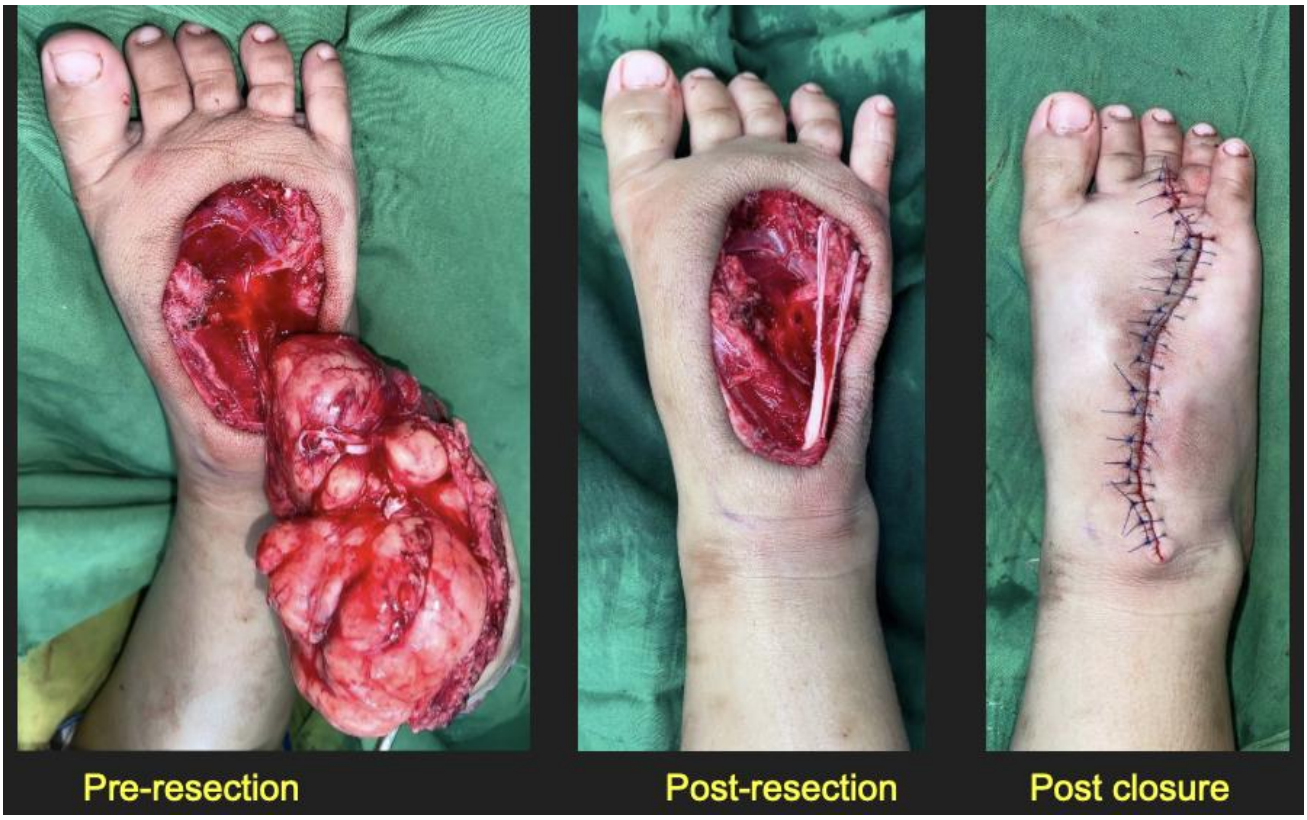

Fig. 6 Intra-operative image of the mass exhibits well-defined, encapsulated nodules, varying colors ranging from yellow to red are observed, indicative of varying proportions of adipose and vascular elements, alongside the presence of large, thick-walled vessels.

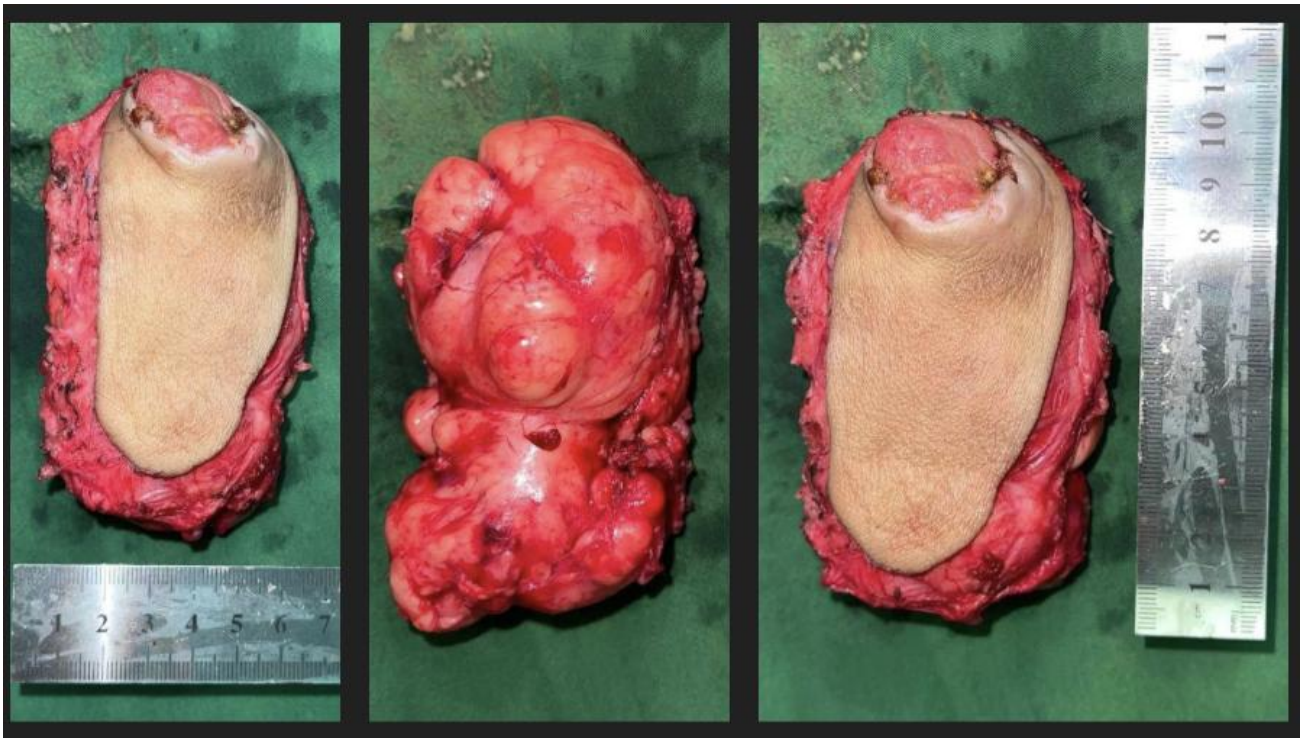
Fig. 7 The removed mass with dimension of 10.5 x 5 cm x 5cm in size and weight 48 grams.

The patient's postoperative recovery was uneventful, and there was no local recurrence noted during the 5 month follow-up period.

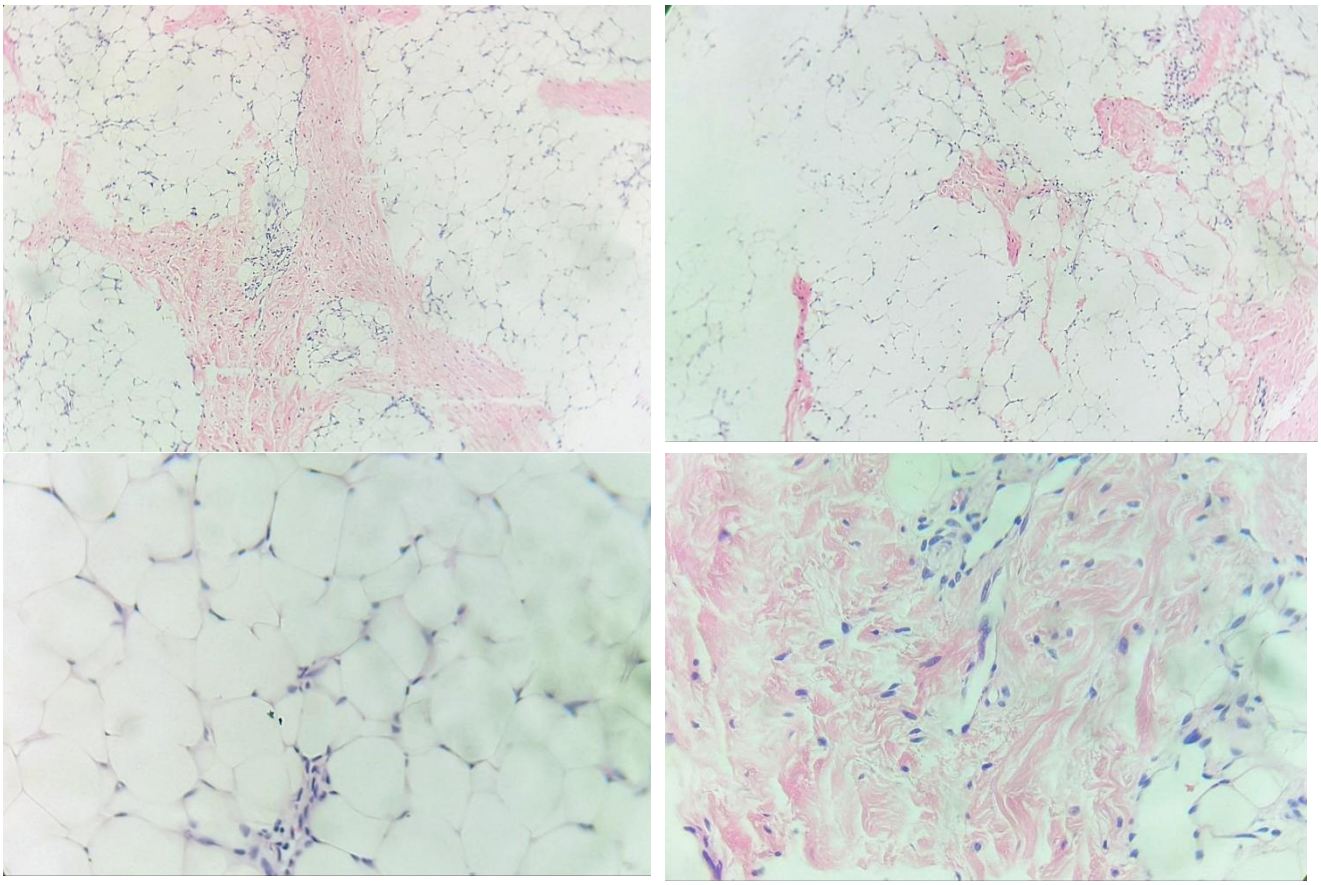

Fig. 8 The histopathological slides reveal mature adipose tissue with small capillary-sized vessels, featuring thin walls and containing bright pink fibrin thrombi, along with occasional thin fibrous septae.

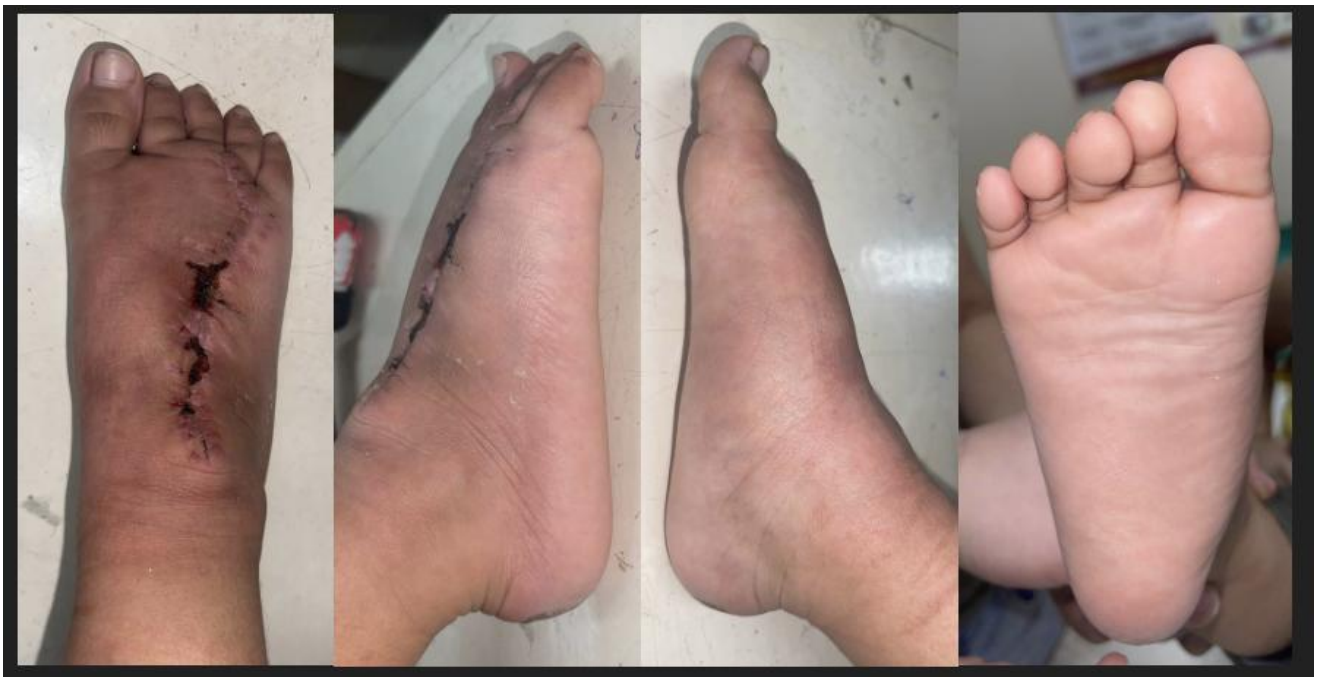

Fig. 9 One month post-surgery, the foot shows a gross image of a dry, well-healed wound with no signs of soft tissue mass recurrence.

Discussion

Angiolipomas are rare benign tumors that are composed of mature adipose tissue and small capillaries5. Typically, they manifest as painless, clearly outlined, unbroken, soft, rounded, movable masses beneath the skin. On rare occasions, they may exhibit a polyp-like shape. They often become tender to the touch, especially during the early stages of growth6. While angiolipomas can occur in any part of the body, they are exceptionally uncommon in the foot.

Angiolipomas predominantly affect young adults, although infiltrating angiolipomas are typically diagnosed in older individuals. They occur with equal frequency in both sexes and are most commonly found in the extremities, particularly the forearm (in about two-thirds of cases), as well as the trunk, spinal axis, head, and neck. Typically, their size does not exceed 4 cm^3.

In this instance, the diagnosis of angiolipoma was verified through histological examination of the excised tissue. Histologically, angiolipomas are characterized by the presence of mature fat cells intermingled with small capillaries. This differs from lipomas, which consist entirely of mature adipose tissue, and hemangiomas, which consist primarily of blood vessels.

Soft tissue masses in the foot can present a wide range of benign and malignant tumors, leading to a broad differential diagnosis. This includes various lipoma subtypes (such as lipomatosis, myolipoma, chondroid lipoma, hibernoma, atypical lipoma, spindle-cell lipoma pleomorphic lipoma, and lipoblastoma), hemangiomas, as well as benign lesions impacting bone, joints, or tendons (such as intraosseous lipoma, parosteal lipoma, lipoma of joint or tendon sheath, and lipoma arborescens), and the possibility of liposarcoma[3]. Although clinical presentation and radiographic findings can provide valuable clues regarding the underlying pathology, a conclusive diagnosis requires histological analysis of the excised tissue.

Surgical removal remains the cornerstone treatment for angiolipomas, especially for infiltrating types where there's an increased recurrence risk. Preserving surrounding tissues is crucial since these tumors are typically encapsulated and well-defined. The surgical approach is determined by factors like the size of the lesion, its location, and its adherence to nearby structures like bone or soft tissue. In instances where complete removal proves challenging, a more extensive surgical approach may be necessary to ensure thorough excision. Moreover, radiotherapy might be recommended after incomplete excision to reduce the likelihood of recurrence.

Despite being benign, angiolipomas can still recur if not completely removed. It's essential to undergo long-term follow-up to watch for any recurrence or potential malignant changes. However, there have been no documented instances of recurrence or malignant transformation following excision[7]. In this particular case, the patient experienced a smooth recovery after surgery, and during a follow-up at five months post-operation, no signs of recurrence were observed.

Conclusions

Angiolipomas are uncommon benign soft tissue tumors characterized by mature fat cells along with an increased vascular component, with infiltrating variants uncommonly found in the foot. Among diagnostic modalities, contrast-enhanced MRI stands out as the most effective tool for precise lesion characterization. Histopathological examination remains imperative for definitive diagnosis, with surgical excision being the preferred treatment approach. The case we've documented presents a unique aspect in terms of both its anatomical location and the patient's demographic characteristics, contributing valuable insights to the existing literature on infiltrating angiolipomas. It's crucial for physicians and surgeons to be vigilant about the potential occurrence of such soft tissue masses and to accurately identify and manage them accordingly.

Additional Information

Disclosures

Human subjects: The participant in this report is provided with a written informed consent.

Conflicts of interest:

Payment/services: The authors confirm that no financial support was received from any organization for the submitted work.

Financial relationships: The authors affirm that they have no financial relationships at present or within the previous three years with any organizations that could have an interest in the submitted work.

Other relationships: The authors assert that there are no other relationships or activities that could be perceived to have influenced the submitted work.

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