



Teaching cases

Infiltrating hybrid mesenchymal tumor of skeletal muscle showing lipomatous, hemangiomatous, leiomyomatous and osseous features – An unusual soft tissue tumor providing insight into the pathogenesis of lipoma variants



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ABSTRACT

Infiltrating angiolipoma and osteolipoma of the hand are rare. A 40-year-old man presented with slowly enlarging swelling of his right hand for two and half years without functional deficit but it became painful with slight limitation of movement for the past few months. Plain radiograph showed a large soft tissue swelling with specks of calcifications. Ultrasonography and magnetic resonance imaging revealed an infiltrative mass in the right palm deep to the flexor tendons. As biopsy suggested infiltrative angiolipoma of skeletal muscle, debulking of the tumor was performed. The tumor showed coexistence of all four mesenchymal elements, fat, blood vessel, smooth muscle and bone in various combinations in the form of angiolipomatous, osteolipomatous, ossified intramuscular hemangiomatous, myolipomatous and angiomyolipomatous patterns throughout the entire tumor. Small meningotheial-like whorls of spindle cells were focally seen, some showed intramembranous ossification forming small woven bony spicules in their centers. There were no atypical cells or lipoblasts. Staining for CDK4, MDM2, p16, HMB45 and Melan A was negative. The diagnosis was “infiltrating hybrid mesenchymal tumor of skeletal muscle showing lipomatous, hemangiomatous, leiomyomatous and osseous features”. The fairly even admixture of the various components supports the neoplastic participation of each individual element. Hybrid mesenchymal tumor most probably originates from multipotent neoplastic cells showing multidirectional differentiation.

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Introduction

Benign lipomatous tumor is the commonest soft tissue mesenchymal neoplasm, finding its occurrence not only in the subcutis but also in virtually every organ [1]. Its pure form lipoma occurs predominantly in the subcutis on the posterior neck, trunk, extremities and axilla. It can additionally harbor other mesenchymal elements as variants of lipoma. Examples include angiolipoma, myolipoma, chondrolipoma and osteolipoma in which proliferative blood vessels, smooth muscle, cartilage and bone respectively are present in a significant amount in the neoplasm [1]. Furthermore, in addition to its usual thinly encapsulated form, it can assume an infiltrative character as infiltrating lipoma and infiltrating angiolipoma

typically found intramuscularly in the upper or lower extremities [2].

Infiltrating angiolipoma and osteolipoma of the hand are both rare with only few cases reported in the literature [3,4]. We describe in this article a unique case of infiltrating hybrid mesenchymal tumor of the hand consisting of an angiolipomatous background in which are numerous evenly distributed lamellar bone fragments and focal meningotheial-like whorls of spindle cells, some showing intramembranous ossification. In addition, aggregates of interlacing smooth muscle fascicles are also seen scattered throughout the tumor. Its implication in the understanding of the pathogenesis and nomenclature of lipoma variants are discussed.

Clinical history

A 40-year-old man presented with slowly enlarging swelling of his right hand for two and half years without much functional deficit. For the past few months, it had become slightly painful and affected his finger movement. He recalled no preceding trauma or

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fever. Physical examination revealed diffuse swelling of his right hand, especially over the first web space but also extending into the palmar and dorsal aspects. There was mild tenderness but no redness or hotness. Plain radiograph showed a huge soft tissue swelling with specks of calcifications, and periosteal reaction over third and fourth metacarpal shafts (Fig. 1A). Ultrasonography revealed a large tumor deep to the flexor digitorum profundus with dorsal extension into the interosseous muscles. There was moderate internal vascularity with multiple calcifications. Magnetic resonance imaging (MRI) disclosed a 7.6 cm × 6.5 cm × 4.2 cm contrast enhanced mass with main bulk in the right palm deep to the flexor tendons of the thumb and fingers and dorsal extension through the Interosseous muscles (Fig. 1B). In view of the history of recent increase in size, periosteal thickening of the third and fourth metacarpal and multiple calcifications, the differential diagnosis included synovial giant cell tumor and synovial sarcoma.

Since ultrasound-guided biopsy of the mass revealed features suggestive of infiltrating angiolipoma of skeletal muscle and complete removal would entail right hand amputation, debulking of the tumor was performed for maximal functional restoration. The operative findings were those of a mixed adipose and vascular lesion with prominent spotty calcification, suggestive of an infiltrating angiolipoma with phlebolith formation. Apart from some weakness in the extension of middle and ring fingers, and diminished sensation over the first web space, the patient regained almost full hand function and remained well 13 months after operation.

Materials and method

The biopsy and surgically removed tissues were fixed in 10% buffered formalin and processed for routine histological analyses. For immunohistochemical studies, the avidin–biotin complex was employed as a detection system, with microwave antigen retrieval prior to applying the primary antibodies. The Ventana Ultraview detection kit and Benchmark XT automated immunostaining system were used.

Result

Pathological findings of the biopsy

The small biopsy fragments consisted of skeletal muscle bundles infiltrated by lobules of mature adipocytes in which were scattered blood vessels of varying sizes with surrounding mild infiltrates of small lymphocytes. The features were indicative of infiltrating angiolipoma of skeletal muscle.

Pathological findings of the removed tumor

The removed tumors comprised three nodular masses, 4 cm × 3 cm × 3 cm, 3 cm × 3 cm × 2 cm and 2 cm × 2 cm × 2 cm across. Their cut surfaces showed lobulated yellow fatty tissues in which were scattered dark brown spongy nodules of varying sizes (Fig. 2A). In the edges of some of these spongy nodules were yellow specks with a gritty sensation on sectioning, suggestive of calcified or bony tissue (Fig. 2A).

Histologically, the tumor infiltrated into surrounding skeletal muscles. The fatty component consisted of lobules of mature adipocytes in which were arborizing blood vessels of varying sizes; occasional fibrin microthrombi were seen in some of the small-size vessels (Fig. 2B). The spongy areas were composed of aggregates of dilated venous channels imparting a cavernous hemangiomatic appearance (Fig. 2C). In the borders of many of these venous

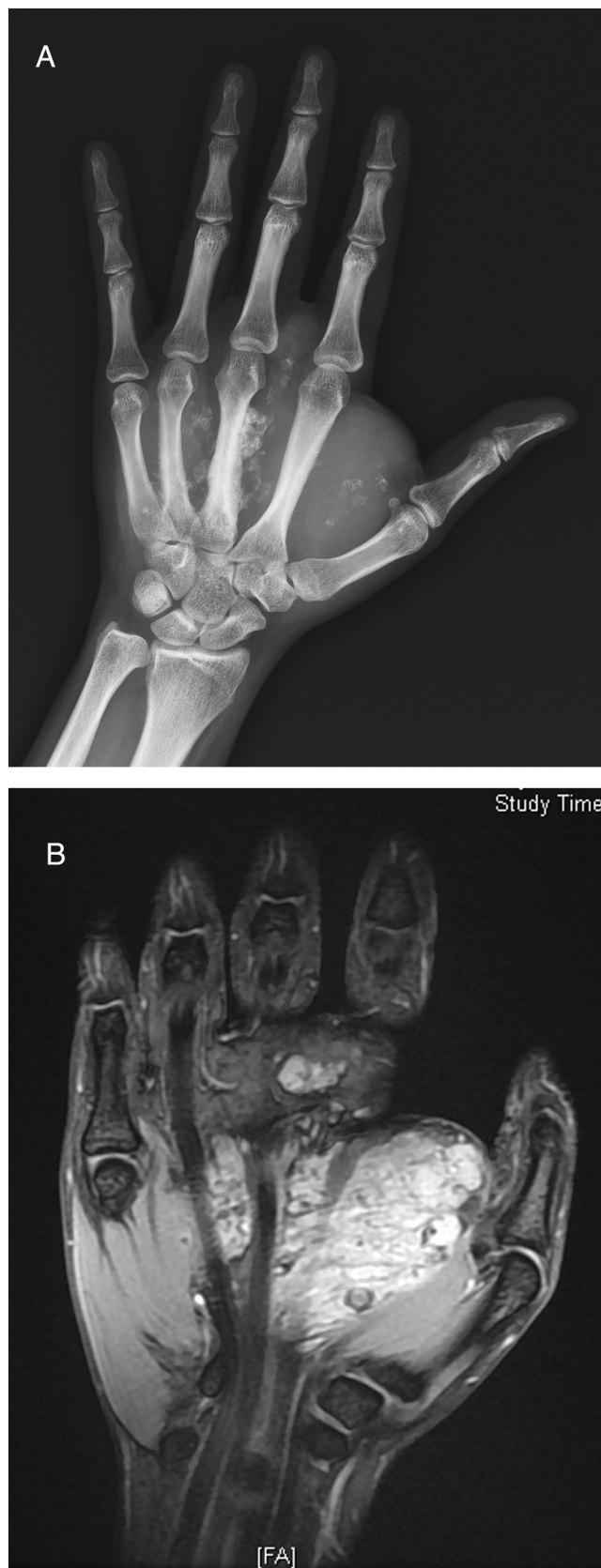


Fig. 1. (A) Plain radiograph shows a huge soft tissue swelling with specks of calcifications, and periosteal reaction over third and fourth metacarpal shafts. (B) MRI reveals a large contrast enhanced mass with main bulk in the right palm deep to the flexor tendons of the thumb and fingers and dorsal extension through the interosseous muscles (proton-density with fat saturation sequence).

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