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Intraosseous neurilemmoma of the proximal ulna

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ABSTRACT

INTRODUCTION: Neurilemmoma is a benign nerve sheath neoplasm commonly located in the soft tissue. Intraosseous neurilemmoma is rare, constituting less than 1% of primary bone tumors.
PRESENTATION OF CASE: A 21 year-old woman was presented with left elbow pain of 1-month duration. Plain radiographs showed a well-defined, lytic and expansile lesion of the proximal ulna. Computed tomography revealed cortical destruction and soft tissue extension. Because the tissue of origin for the tumor was uncertain, an open biopsy was performed. The specimens demonstrated a benign spindle cell tumor suggestive of a neurilemmoma, similar to a soft tissue neurilemmoma. The diagnosis of intraosseous neurilemmoma was established. Marginal excision of the soft tissue component and curettage of the lesion in the bone were performed. After 3.5 years of follow up, there is no clinical or radiographic finding to suggest any recurrence.
DISCUSSION: The major site of intraosseous neurilemmoma is the mandible. Occurrence in the long bone is particularly rare. Only two cases of intraosseous neurilemmoma involving the bones around the elbow have been reported to our knowledge; these cases arose in the distal humerus. We describe the first case of intraosseous neurilemmoma of the proximal ulna of the left elbow. The recommended treatment is conservative resection and bone grafting, as malignant change is extremely rare.
CONCLUSION: Although very rare, intraosseous neurilemmoma should be taken under consideration in the differential diagnosis of painful, radiographically benign-appearing osseous tumor around the elbow.

1. Introduction

Neurilemmoma (schwannoma) is a benign neoplasm that arises from the myelinating schwann cells of the nerve sheath.[1–3] It is a relatively common tumor, comprising approximately 5% of all benign soft tissue tumors, and shows a predilection to affect the sensory nerves.[3] Intraosseous neurilemmoma, however, is very rare, accounting for less than 1% of benign bone tumors.[4,5] It shows no sex-, race-, or age-dependant predilections.[6] The mandible is the most frequently affected site, followed by the sacrum; this tumor rarely arises in the bones of the extremities.[5,7,8] Only 2 cases of intraosseous neurilemmoma around the elbow have been described in literature[6,9]; these cases arose in the distal humerus. We present the first case report of an intraosseous neurilemmoma affecting the proximal ulna.

2. Presentation of case

A 21-year-old woman was referred to our orthopedic unit with a 1-month history of pain in the left elbow. The initiating factor, such as trauma, was not clear. On physical examination, the overlying skin was intact, and there was no evidence of warmth, erythema, or induration. No other masses were palpable. The elbows and forearms had a normal range of motion (ROM). No evidence of lymphadenopathy or neurovascular involvement was found.

Plain radiographs revealed a well-defined, lytic, and expansile lesion, with thin marginal sclerosis and trabeculation in the proximal ulna (Fig. 1). Computed tomography (CT) showed considerable destruction of the cortex of the ulna, which connected with a mass of soft tissue. At the edge of the destructed cortex overhung a soft tissue mass (Fig. 2a), which had invaded into the cortex, and no periosteal reaction was seen (Fig. 2b). Magnetic resonance imaging (MRI) showed the well-defined and lobulated lesion to be isointense to skeletal muscle on T1-weighted images, and heterogeneous and hyperintense on T2-weighted images. The lesion revealed uniform enhancement following Gd-DTPA administration.

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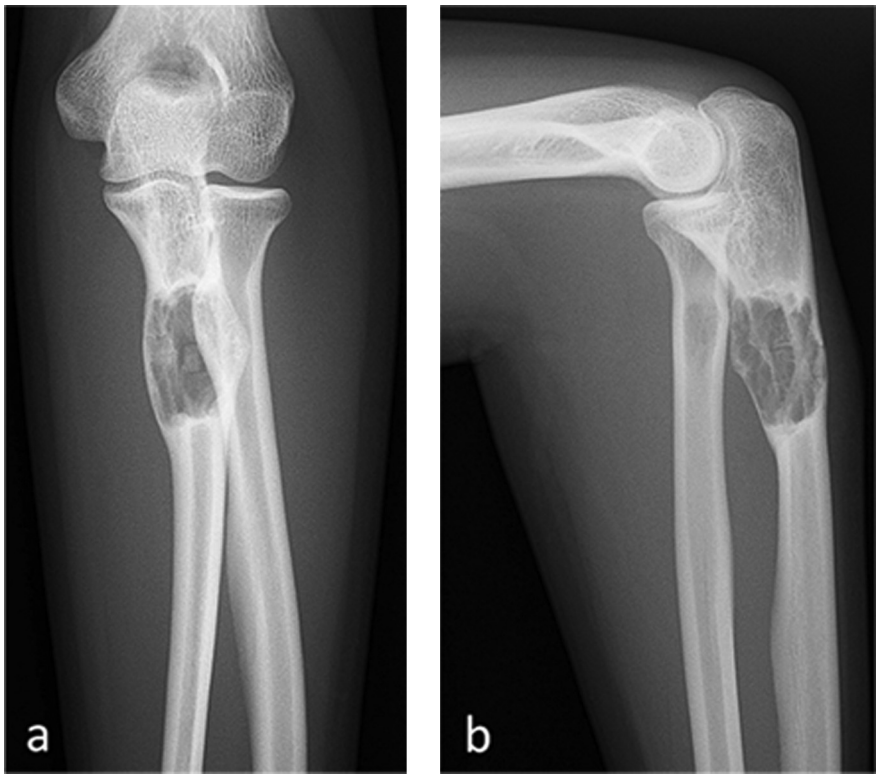

Fig. 1. Anteroposterior (a) and lateral (b) plain radiographs, showing a well-defined, lytic, and expansile lesion, with marginal thin sclerosis and trabeculation in the proximal ulna.

Because of uncertainty regarding the histological origin of the tumor, we performed an open biopsy. The pathologic specimen consisted of hypercellular regions with palisading nuclei (Antoni type A), and hypocellularity regions with myxoid background (Antoni type B). Almost all of the area was Antoni type A (Fig. 3). We could not observe nuclear atypia, necrosis, or mitosis. Immunohistochemical studies showed strong, diffuse reactivity for S-100 protein within the lesional cells. Microscopic findings were consistent with a diagnosis of neurilemmoma.

The diagnosis of intraosseous neurilemmoma was established. We surgically excised the soft tissue component of the mass by marginal resection. Curettage was performed on the bone lesion, and the resulting deficit was grafted with beta-tricalcium

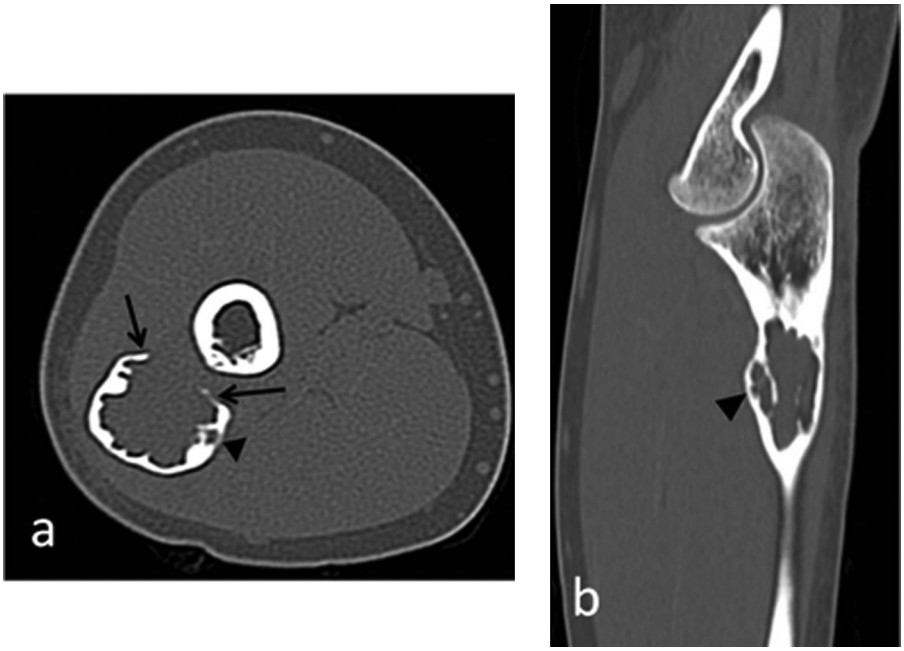

Fig. 2. Axial computed tomography scan (a) and sagittal reconstruction (b) of the ulna, demonstrating destruction of the cortex of the ulna, with extension into the adjacent soft tissue. At the edge of the destructed cortex overhung a soft tissue mass (arrows), which had invaded into the cortex (arrowhead).