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CASE REPORT

Intraosseous schwannoma of the olecranon: A case report

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ABSTRACT

Intraosseous schwannomas are extremely rare benign tumors originating from Schwann cells of the peripheral nervous system. While these tumors are commonly found in the mandible and sacrum, their occurrence in long bones, particularly the ulna, is uncommon. A 59-year-old female patient was admitted with a two-year history of pain and swelling in her right elbow. Radiographic imaging revealed a lytic lesion with cortical expansion and thinning in the proximal ulna metaphysis, while magnetic resonance imaging showed a lesion with irregular borders extending into the joint via cortical erosion. Differential diagnoses included a simple bone cyst, enchondroma, giant cell tumor, and metastasis. Surgical procedure involved curettage and bone grafting, and the lesion was diagnosed histologically as an intraosseous schwannoma. Immunohistochemical staining showed positive expression of SOX10 and S100, confirming the diagnosis. Postoperative recovery was uneventful, and the patient regained a full range of motion without pain at her six-month follow-up. In conclusion, this case underscores the importance of considering intraosseous schwannoma in the differential diagnosis of benign bone lesions, particularly in rare locations such as the ulna. Histopathological examination remains essential for an accurate diagnosis.

Keywords: Bone neoplasms, intraosseous schwannoma, olecranon, rare tumors, ulna bone.

Schwannomas are benign tumors derived from Schwann cells, which form the myelin sheath of the peripheral nerve cells.[1-3] They are among the most common tumors of the nervous system; however, intraosseous schwannomas account for less than 0.2% of primary bone tumors.[1,4,5] While intraosseous schwannomas typically affect the mandible and sacrum, involvement of long bones is uncommon.[6-8] There is no significant tendency based on sex or ethnicity.[9] They can occur at any age, although they are most commonly observed in individuals aged 40 to 60 years.[10]

The proposed mechanism for their formation suggests that as nerve fibers traverse the bony canals along with blood vessels, intraosseous formation may occur.[8] The non-specific nature of the clinical presentation and the potential for misdiagnosis with other tumors which exhibit similar radiographic findings make accurate preliminary diagnosis of intraosseous schwannomas challenging.[1] Various treatment options have been documented in the literature, including surgical resection, curettage, autogenous and allogenous bone grafting, and cement filling techniques.[4,6]

Approximately 20 cases of intraosseous schwannomas in long bones have been reported in the literature,[7] with only three cases located in the ulna.[4,8,9] In this article, we present a case of intraosseous schwannoma of the olecranon presenting as a primary bone tumor.

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A 59-year-old female patient was admitted with no known chronic illnesses. Her medical history and

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family history were unremarkable for hereditary diseases such as neurofibromatosis. She was referred to our clinic from an external center after presenting with a two-year history of pain and swelling in her right elbow. There was no history of trauma or infection. Upon physical examination, the range of motion of the elbow was intact, although tenderness was noted with palpation, and pain increased with movement. No neurovascular pathology was observed in the extremity, and laboratory results were normal. X-rays revealed a lytic lesion in the right proximal ulna metaphysis with lobulated features, cortical bone expansion and thinning, characterized by a sclerotic rim (Figure 1). No significant calcification was observed. Magnetic resonance imaging (MRI) showed an intraosseous, irregularly bordered, contrast enhanced lesion measuring 8.1×4.3×4.1 cm in the metaphysis of the right ulna, extending into the joint via cortical erosion (Figure 2). There was no periosteal reaction or soft tissue extension. The lesion appeared isointense to muscle tissue on T1-weighted images and locally heterogeneously hyperintense on T2-weighted images. Based on imaging findings and clinical symptoms, differential diagnoses included simple bone cyst, enchondroma, giant cell tumor, and metastasis. Surgical procedure was planned as curettage and grafting.

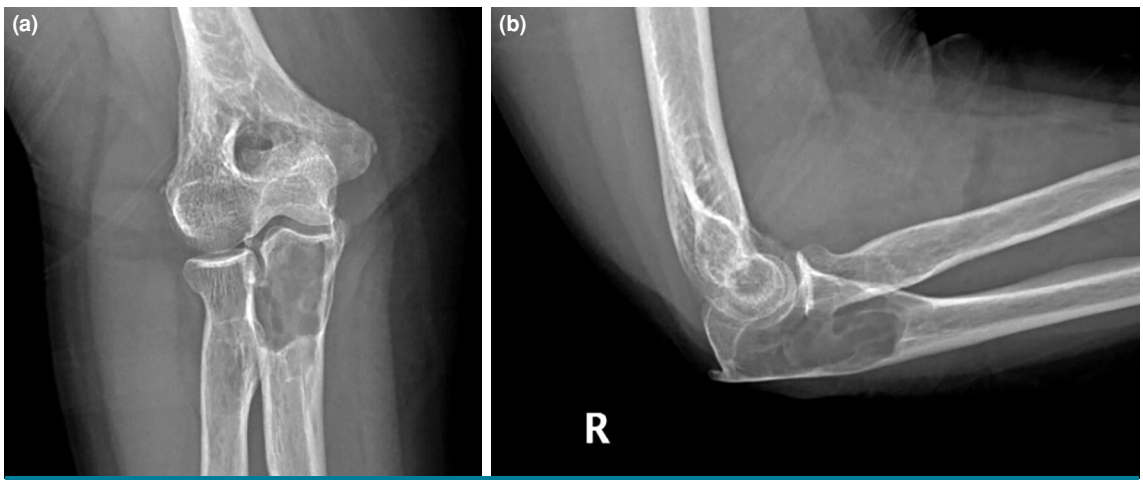


FIGURE 1. Preoperative X-ray appearance. **(a)** Anteroposterior X-ray. **(b)** Lateral X-ray.

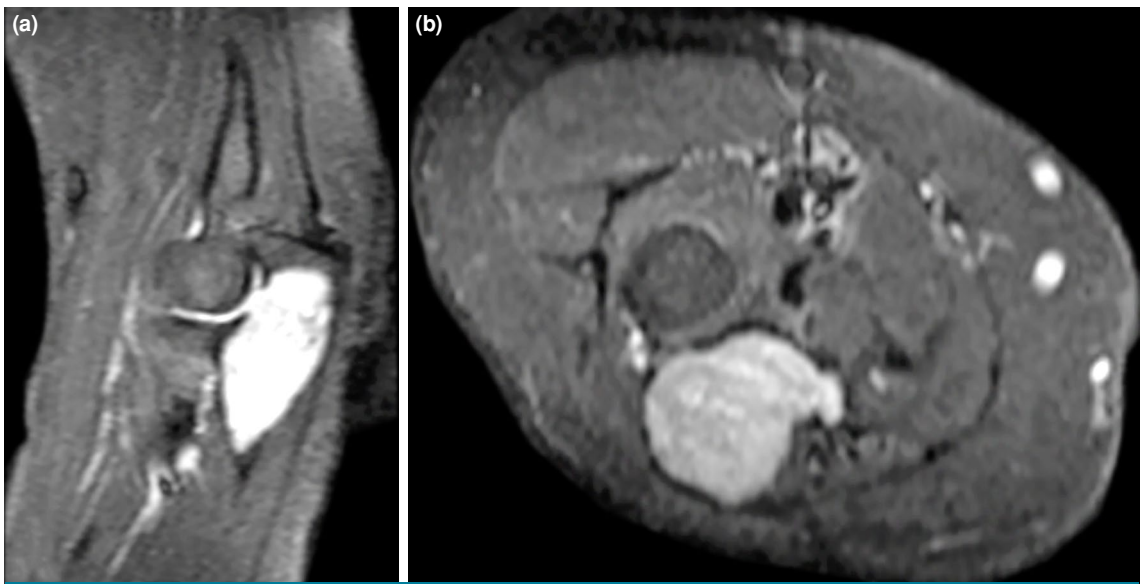


FIGURE 2. Preoperative magnetic resonance imaging appearance. T2-weighted images. **(a)** sagittal section **(b)** axial section.

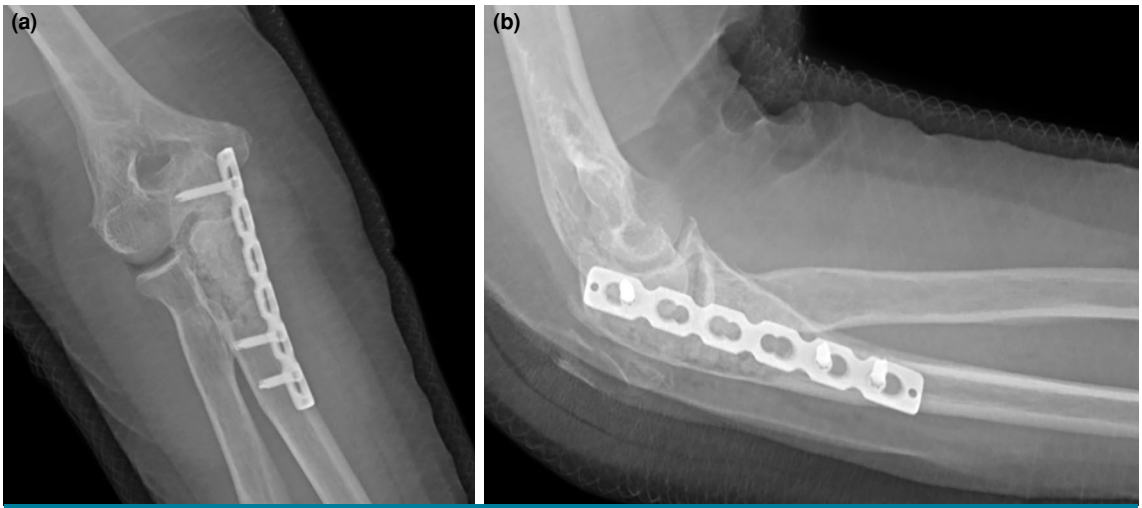


FIGURE 3. Postoperative X-ray appearance. **(a)** Anteroposterior X-ray **(b)** Lateral X-ray.

After performing a supraclavicular block on the patient's right upper extremity, a tourniquet was applied. A 10-cm skin incision was made over the olecranon in the supine position. A 4×2 cm cap-like section of the weakened cortex was elevated at the medial aspect of the proximal ulna, where the bone was slightly expanded. The mass was carefully curetted and excised for pathology. After thorough irrigation, a 1.5×1 cm cortical defect was observed on the joint-facing surface of the proximal ulna, with the trochlear cartilage surface visible. A cartilage matrix scaffold was placed at the defect site, and the medulla was filled with human-derived bone allograft and impacted. Under fluoroscopic guidance, the cap was repositioned, and a 3.5-mm reconstruction locking plate was placed (Figure 3). No intraoperative complications occurred. The patient was discharged the day after surgery. The right elbow was immobilized with a long arm splint for two weeks, after which active movement was initiated.

The curettage material comprised 6 mL of tan-colored, firm tissue fragments. On microscopic

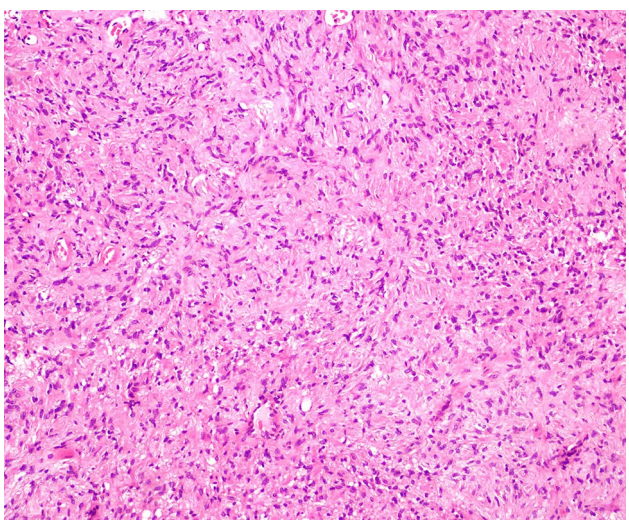


FIGURE 4. Haphazardly arranged spindle cells in hyalinized stroma (H&E, ×100).

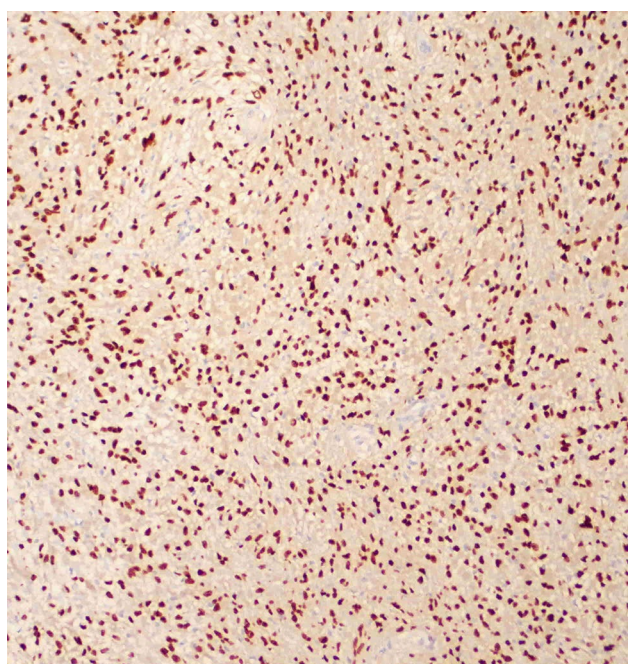


FIGURE 5. Diffuse nuclear SOX10 expression in tumor cells (SOX10 ×100).

TABLE I
Summary of all published cases of ulna intraosseous schwannomas

Number	Authors	Published year	Surgical options
1	Kito et al.[9]	2014	Curettage, beta-tricalcium phosphate graft
2	Suzuki et al.[8]	2016	Curettage, hydroxyapatite granule, plate fixation
3	Lim and Wu[4]	2021	Curettage, hydroxyapatite and beta-tricalcium phosphate graft, bone cement, plate fixation

examination, hematoxylin-eosin (HE)-stained sections showed an unencapsulated tumor composing of compact sheets and fascicules of spindle cells accompanied by loosely arranged foci, hyalinization and cystic degeneration (Figure 4). There were lipid laden histiocytes and thick-walled hyalinized blood vessels in some areas. Tumor cells were bland spindle cells with undulating fusiform cytoplasm. Small intranuclear inclusions were readily observed in tumor cells. Hypercellularity, necrosis, nuclear atypia or increased mitosis were not observed. Morphological differential diagnosis included schwannoma, neurofibroma, leiomyoma/leiomyosarcoma, desmoplastic fibroma and to a lesser extent metastatic sarcomatous carcinoma. In immunohistochemical tests, tumor cells expressed diffuse nuclear SOX10 and S100 positivity (Figure 5), while actin, desmin, pancytokeratin and CD34 were negative. Based on morphological and immunohistochemical features, a diagnosis of intraosseous schwannoma was made.

During follow-up, the patient experienced no complications, and at the six-month follow-up, the elbow range of motion was full and pain-free. A written informed consent was obtained from the patient.

DISCUSSION

Schwannomas are slow-growing benign peripheral nerve tumors originating from Schwann cells, which are responsible for production the myelin sheath surrounding peripheral nerve cells.[11] Intraosseous appearance of schwannomas is quite rare. Gordon[12] reported that most intraosseous nerves such as the radial or fibular nerve are unmyelinated and participate only in vasomotor functions, which is different from the mandibular nerve originating from the trigeminal nerve.[13] Intraosseous schwannomas are most frequently seen in the mandible and sacrum, where sensory nerves are predominantly located, but their appearance in long bones is extremely rare.[3,9] Three approaches have been reported regarding intraosseous involvement of schwannomas:[9,14] *(i)* tumors originating centrally within the bone, *(ii)* tumors developing in the nutrient foramen, and *(iii)* secondary bone erosion due to an extraosseous tumor. In the anatomy of the proximal ulna, the fact that the branches of the nutrient arteries enter the anterior cortex of the metaphyseal region 7.5 cm distal from the tip of the olecranon and course intraosseously supports the current mechanism.[15]

Most of these tumors are diagnosed incidentally and the pain has an insidious onset in the clinic.[5] Pathological fractures may occur following trauma. Our case involved insidious onset with aggravating pain upon movement over a two-year period.

On radiographic images, intraosseous schwannoma appears as lytic lesions characterized by a sclerotic rim with cortical trabeculation and erosion.[3,14,16] It typically does not contain intralesional calcification.[1,9] According to Knight et al.,[17] MRI is essential, as radiographic images do not effectively distinguish between benign and malignant lesions. On MRI, schwannomas typically appear as heterogeneous masses which are isointense to skeletal muscle on T1-weighted images and hyperintense on T2-weighted images.[1,3,16] Although these radiographic descriptions are consistent with our patient's findings, they are not specific for schwannoma.

Imaging and clinical data may suggest other benign primary bone tumors such as simple cyst, aneurysmal bone cyst, giant cell tumor, enchondroma, fibrous dysplasia, chondromyxoid fibroma, or neurofibroma. Primary malignant bone tumors usually grow more rapidly, but care should be taken to exclude these tumors before pathology results are known.[11,14] Histopathological analysis is indispensable for accurate diagnosis. In our case, after evaluating imaging and clinical course, we suspected a benign bone tumor and did not perform a biopsy. The intraoperatively curetted lesion was sent for histopathological analysis.

Schwannomas have a wide range of histomorphology. The pathognomonic histological pattern has been described as a biphasic histological pattern with Antoni A and B areas. Antoni A areas are hypercellular areas and consist of spindle cells and their nuclei can be arranged in palisades forming Verocay bodies.[18] Antoni B areas are hypocellular areas arranged in the myxoid stroma.[5] Unfortunately, these pathognomonic patterns are only rarely seen in conventional schwannomas that and our case did not show either of these features, either. According to Flores Santos et al.[16] and Haberal et al.,[3] schwannomas show intense positivity for S-100 protein, which serves as diagnostic confirmation and makes it possible to distinguish schwannomas from lesions such as fibrosarcoma, fibrous dysplasia, and non-ossifying fibroma. Karamchandani et al.[19] reported that SOX10 was expressed more than S-100 in peripheral nerve sheath tumors, particularly in intraosseous schwannoma. The SOX10 expression was also demonstrated in our case.

Various treatment options have been described in the literature. The treatment for intraosseous schwannoma typically involves curettage followed by placement of autogenous or allogenous bone graft.[1,3,17] Autograft, particularly iliac crest bone graft, is the most commonly used, but significant donor site morbidity may occur. There are cases in which bone cement is applied. Lim and Wu[4] showed that cement increased stability in osteoporotic bone and provides an antineoplastic effect against residual tumor cells through hyperthermia. Since cortex thinning or formation of a large defect at the end of curettage may cause pathological fracture, stabilization with osteosynthesis may be applied.[4] No malignant transformation has been reported.[16] The reason for recurrence in cases reported in the literature is inadequate curettage or excision of the lesion.[3,20] In the literature, three cases of intraosseous schwannoma in the proximal ulna have been reported, each with different treatment approaches and follow-up durations (Table I). In our case, we performed meticulous curettage, used an allogeneic bone graft, and provided additional stabilization with plate fixation. Of note, this combined approach, involving cartilage matrix scaffold, human-derived bone allograft, and plate fixation, differs from previous cases in the literature. The patient's follow-up is still ongoing, and the unique aspects of the case contribute to the current understanding of intraosseous schwannomas.

In conclusion, intraosseous schwannoma is an extremely rare tumor, making accurate pre-diagnosis quite challenging. Therefore, histopathological analysis is crucial. This case emphasizes that intraosseous schwannoma should be considered in the differential diagnosis of patients presenting with lesions in long bones that appear benign on radiological imaging.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept: Ş.Ç.; Design, literature review, writing the article: Ö.F.N.; Control/supervision, critical review: S.G., Ş.G.; Data collection and/or processing: N.A.; Analysis and/or interpretation: Ş.G., N.A.

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