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Case Report

A Rare Case of Intraosseous Spindle Cell Hemangioendothelioma in the Ischium

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ABSTRACT

Background: Spindle cell hemangioendothelioma (SCH) is an uncommon vascular tumor that rarely affects bone tissue.

Objectives: This report describes a unique intraosseous SCH of the ischium.

Case Presentation: A 55-year-old woman presented with the chief complaint of pelvic pain, mainly on the right side. Radiography and computed tomography (CT) revealed a lytic lesion on the right ischium. CT-guided biopsy revealed a spindle cell area without malignant components. The patient underwent curettage and bone graft surgery, and the pathology report indicated spindle cell hemangioendothelioma.

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Introduction

Spindle cell hemangioendothelioma (SCH) is a rare benign tumor characterized by thin-walled, cavernous, and vascular spaces. Some spaces contain phleboliths, while others are filled with cellular stroma composed of spindled fibroblastic cells. Weiss and Enzinger were the first to mention this in 1986 [1]. Although initially considered a moderately malignant vascular tumor due to its tendency for local recurrence, SCH's malignant potential was later re-evaluated and found to have a low metastatic potential. This type of tumor is most commonly found in soft tissues, but its occurrence in bone tissues is infrequent [2]. Typically, spindle cell hemangiomas are superficial or subcutaneous erythematous nodules that tend to appear in the limbs of young individuals, particularly the hands, with an equal sex distribution [3]. SCH pathologic features include cavernous blood vessels and solid regions of spindle and epithelioid cells [4]. It was formerly assumed to be a tumor with limited metastatic potential, but it is now classified as a benign vascular tumor rather than a low-grade angiosarcoma [5].

This article aims to introduce a known case of intraosseous SCH lesion in a 55-year-old woman in the ischium bone who presented with pelvic pain.

Case Presentation

A 55-year-old woman presented with pelvic pain that was mostly felt on the right side. Pelvic radiography showed a significant decrease in the density of the right ischium (Figure 1). Computed tomography (CT) scan of the ischium indicated a lytic expanding lesion in the ischium (Figure 2). Pathology showed a spindle cell area after the patient underwent a CT-guided biopsy.

Surgical procedure

After the pathological results were determined, a bone curettage surgical plan was arranged for the patient. Following spinal anesthesia, the patient was placed in a prone position with the hips and knees lightly flexed. An incision was made through the gluteus maximus and hamstring muscles down to the ischium. Curettage was performed, and a bone graft was placed.

Pathology

A sample was extracted for histological study, and the results showed an intra-osseous neoplasm composed of two distinct components: Vascular and solid cells. The periphery of the tumor exhibited loose vascular channels intermingled with a centrally located hypercellular area, consisting of monomorphic, bland-looking spindle cell proliferation in septa of vascular spaces (Figure 3).

Follow-up

The patient was followed-up for one year and had no complaints. The last pelvic radiography was performed one year after surgery, and complete healing of the bone tissue was evident (Figure 4).

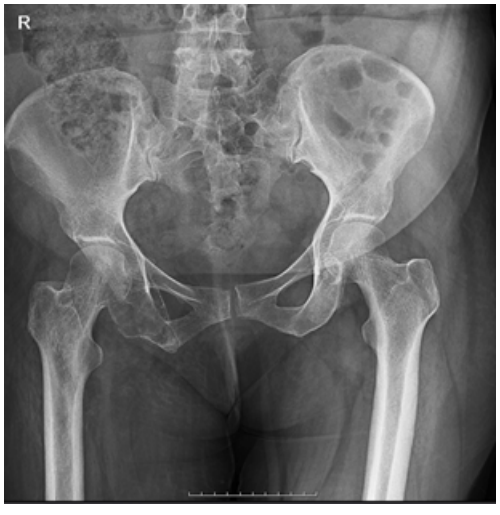

Figure 1. The initial pelvic radiography showing decreased bone density in the right ischium

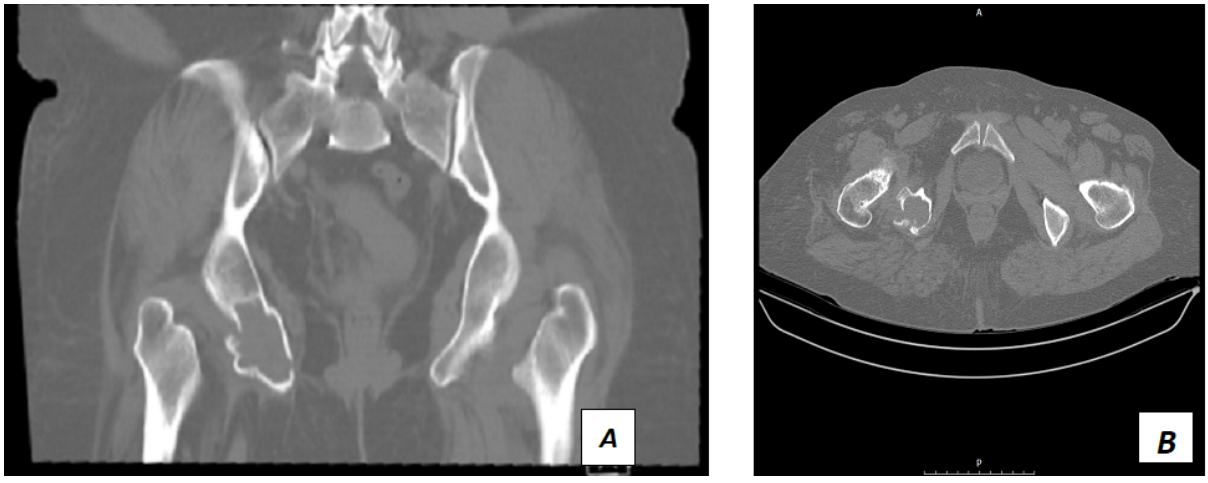

Figure 2. Pelvic CT scan
A) Coronal view, B) Axial view
CT scan: Computed tomography scan.

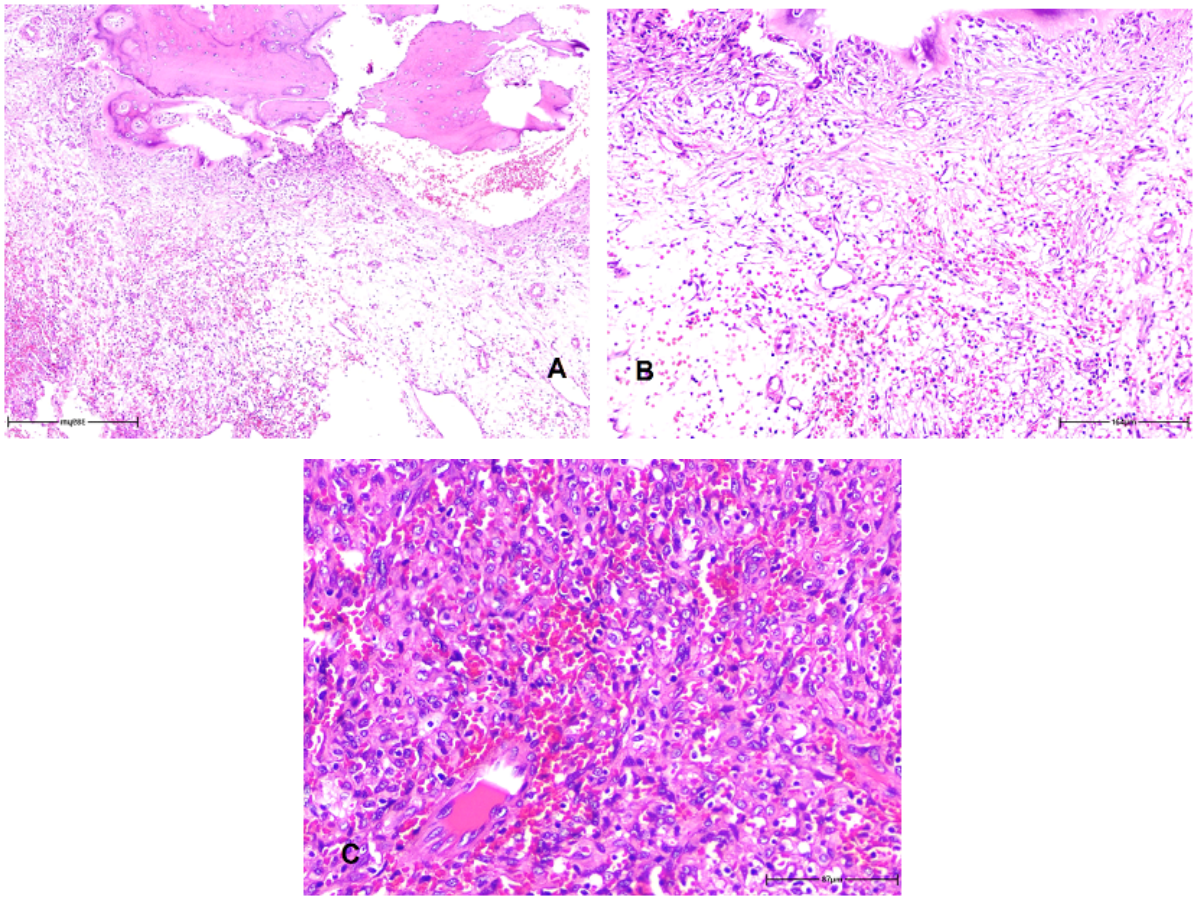

Figure 3. Histopathology sections revealing an intra-osseous neoplasm composed of two distinct components
A) Vascular and solid cells (H&E staining, ×4), B) The periphery of tumor composed of loose vascular channels (H&E staining, ×10), C) Central portions of the neoplasm much more cellular; composed of monomorphic bland looking spindle cell proliferation in septa of vascular spaces (H&E staining, ×40)

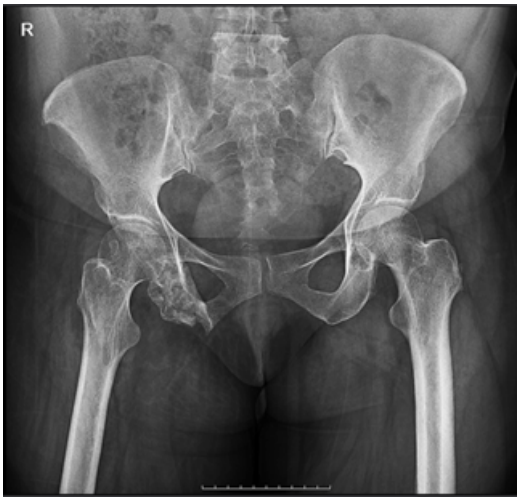

Figure 4. The last radiograph after one year

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Discussion

Tumors originating from bone vasculature are rare, constituting less than 1% of all bone tumors [6]. There are three types of tumors currently classified as epithelioid endothelial cell tumors - epithelioid hemangioma, epithelioid hemangioendothelioma, and epithelioid angiosarcoma [7]. A SCH may also contain a small component of epithelioid endothelial cells. Weiss and Enzinger first described a new type of vascular tumor called "spindle cell hemangioendothelioma" in 1986. This type of tumor has limited malignant potential and is now known as SCH, a benign and often multifocal neoplasm. It typically affects young adults and develops in the subcutaneous tissue of the distal extremities, particularly the hands. In contrast to other hemangiomas, SCH rarely develops in the bone [8]. SCH is histologically characterized as epithelioid and spindle cell hemangioma by the presence of dilated capillaries bordered with flattened endothelial cells filled with red cells, similar to those found in cavernous hemangiomas. These capillaries were tightly mixed with spindle cell components. Furthermore, in SCH, epithelioid cells align in solid cords rather than lining well-formed vascular gaps, similar to epithelioid and spindle cell hemangiomas [2, 9]. Only a few reports of intraosseous spindle cell hemangiomas or hemangioendotheliomas have been published in the medical literature. Spindle cell hemangiomas have been reported in the calcaneus [10], fibula periosteum [11], and frontal bones [12, 13]. Additionally, one case of SCH in the sacrum has been reported [14].

Conclusion

We now describe a rare case of SCH of the ischium. The radiologic differential diagnosis for this case includes several types of tumors, including giant cell tumors of the bone, aneurysmal bone cysts, metastatic bone tumors, solitary bone cysts, and vascular tumors, such as hemangiomas. The patient was managed by surgical curettage and bone grafting; a one-year follow-up showed promising results and was pain-free.

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this research.

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Authors' contributions

All authors contributed equally to the conception and design of the study, data collection and analysis, interception of the results and drafting of the manuscript. Each author approved the final version of the manuscript for submission.

Conflict of interest

The authors declared no conflict of interest.

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