Case Report

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Extraskeletal Osteochondroma in the Posterior Neck of a Middle-Aged Female: A Case Report

중년 여성의 후경부에서 발생한 골격외 골연골종: 증례 보고

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Extraskeletal osteochondroma, a variant of chondroma, typically arises in the para-articular location of hands and feet. It is a rare disease and is particularly uncommon when joint components are not involved or localized away from joints. Herein, we report a case of extraskeletal osteochondroma in the posterior neck of a 66-year-old female. The characteristic radiologic finding of our case is presented, along with the typical findings of the disease and review of related literature reports.

Index terms Osteochondroma; Neck; Middle Age; Adult

INTRODUCTION

Steochondroma is a benign tumor predominantly arising from bone. It is composed of bone and a hyaline cartilage cap, with a connection to the medullary cavity that is continuous with the parent bone. It is a surface lesion that usually arises from the growth plates of the metaphyseal area of the long bones near the joint during the periods of a rapid skeletal growth (1). Rarely, a histologically similar osteocartilaginous lesion can occur in extraosseous structures, and is referred to as extraskeletal osteochondroma. It is a variant of extraskeletal chondroma that has undergone extensive enchondral ossification. Unlike osteochondroma, it is

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not attached to a parent bone. Presenting as a discrete calcified mass, it is commonly reported to be arisen from synovial tissues in joints, tendon sheaths or bursae in the hands and feet (2). It is especially uncommon to be found without involvement of joint components or far away from the joint, and such reported sites include below 1st metatarsal bone shaft, within the thigh, and the nape of neck (3-5). Here, we report a case of extraskeletal osteochondroma developed in the posterior neck, left para-midline area.

CASE REPORT

A 66-year-old female presented with a palpable mass on posterior neck. She had no history of trauma. Physical examination revealed a round to oval shaped, movable, non-tender and protruding mass. There was no neurovascular deficit, and laboratory investigations were unremarkable. Her initial cervical spine lateral radiograph revealed a huge mass with multiple conglomerated ossifications and some calcifications in the soft tissue of posterior neck (Fig. 1A). The lesion showed no medullary or cortical continuity with adjacent bones. On non-enhanced spine CT, approximately 4.4 cm × 2.9 cm × 7.0 cm sized well-marginated heterogeneous but predominantly fat density soft tissue mass was seen in posterior neck from the suboccipital region down to the C5 level, near posterior margin of spinous processes (Fig. 1B). Internally, the mass contained multiple amorphous and conglomerated ossified structures, but no bony continuation to cervical spine was seen. On MR, the mass also demonstrated multiple conglomerated internal ossifications with T1, T2 heterogeneous high signal intensity representing bone marrow and T1, T2 dark signal intensity cortical rimming in a fatty background, with several tortuous signal void vascular structures and subtle heterogeneous en-

Fig. 1. Extraskeletal osteochondroma in the posterior neck of a 66-year-old female.
A. The initial radiograph shows a bony mass in the posterior neck. No connection with the adjacent bony cortex or marrow cavity is observed.
B. Sagittal nonenhanced spine CT image shows a well-marginated heterogeneous soft tissue mass with multiple amorphous ossified structures and fat densities in the posterior neck from the suboccipital region down to the C5 level.

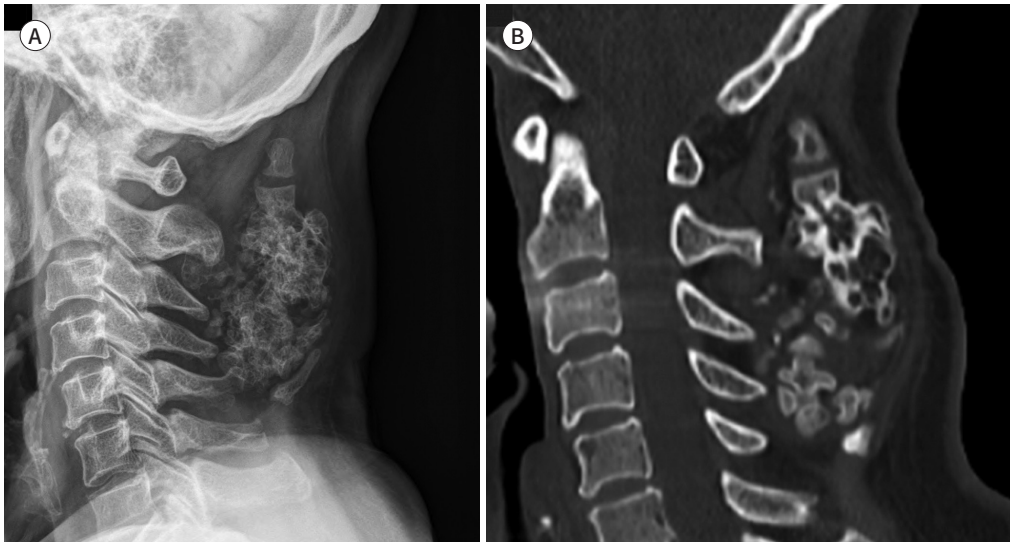
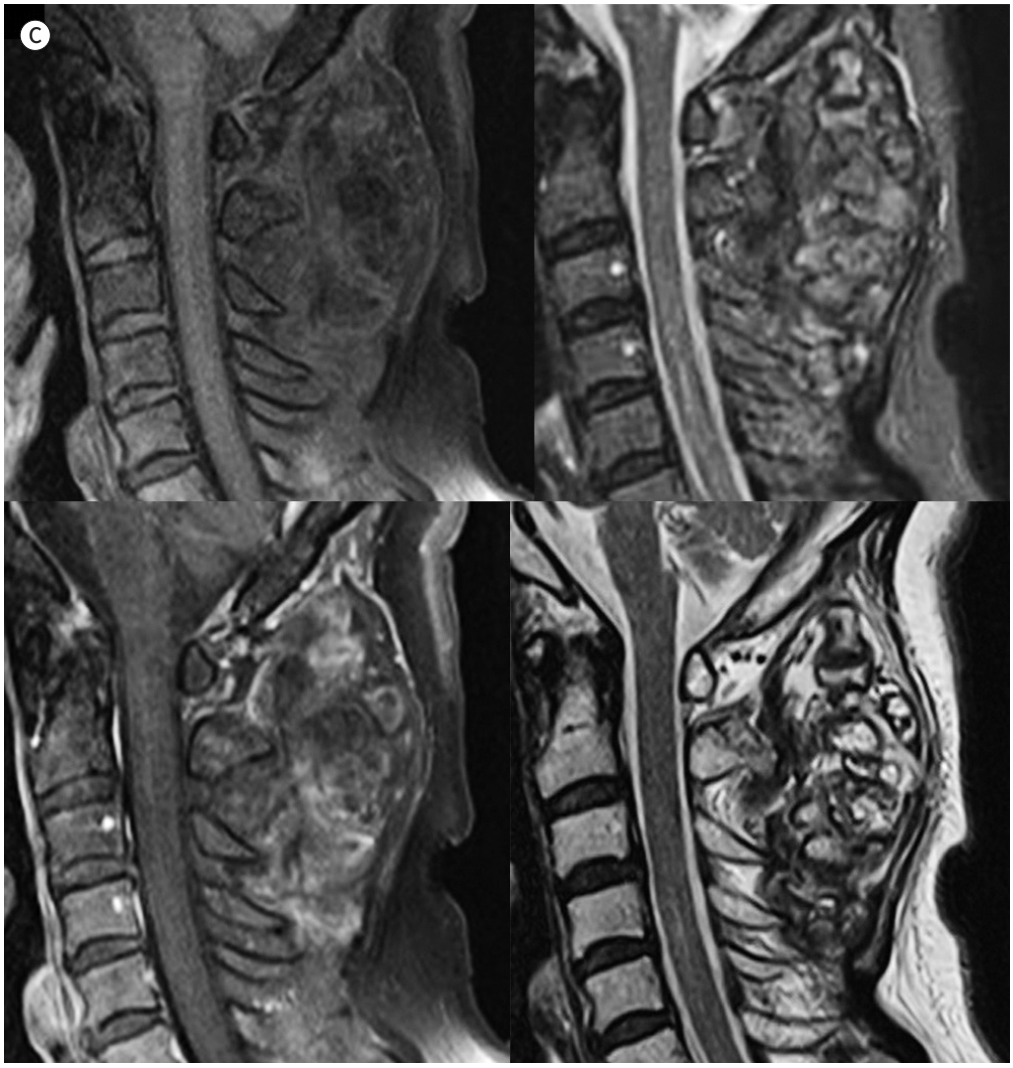


Fig. 1. Extraskeletal osteochondroma in the posterior neck of a 66-year-old female.
C. Sagittal spine MRI images (T1FS, T2FS, T2WI, and FS T1CE in clockwise order) demonstrate the mass with heterogeneous T1 and T2 high signal intensity and subtle heterogeneous enhancement. The mass presents areas of bright signal intensity in conventional T2WI showing dark signal intensity in T2FS images, indicating a fatty background.
FS = fat suppression, T1CE = T1-weighted contrast-enhanced image, T1FS = FS T1-weighted image, T1WI = T1-weighted image, T2FS = FS T2-weighted image, T2WI = T2-weighted image



hancement (Fig. 1C, D). The mass was shifting the adjacent nuchal ligament to right, and mild external pressure erosion at C2 spinous process was seen (Fig. 1D). As differential diagnoses, we considered osteolipoma first accounting for its fatty background. Hemangioma was also considered due to its abundant vascular structures. Finally, we mentioned extraskeletal osteochondroma for our least likely diagnosis.

The patient underwent an excisional biopsy. Intraoperative finding was a well-defined mass that was easily removed, clearly separated from adjacent structures including nuchal ligament. The pathological examination revealed a 7.3 cm × 4.7 cm × 4.0 cm sized solid-fibrotic mass with abundant adipose tissue, cartilage and osteoid components. The histological findings were consistent with those of extraskeletal osteochondroma, demonstrating en-

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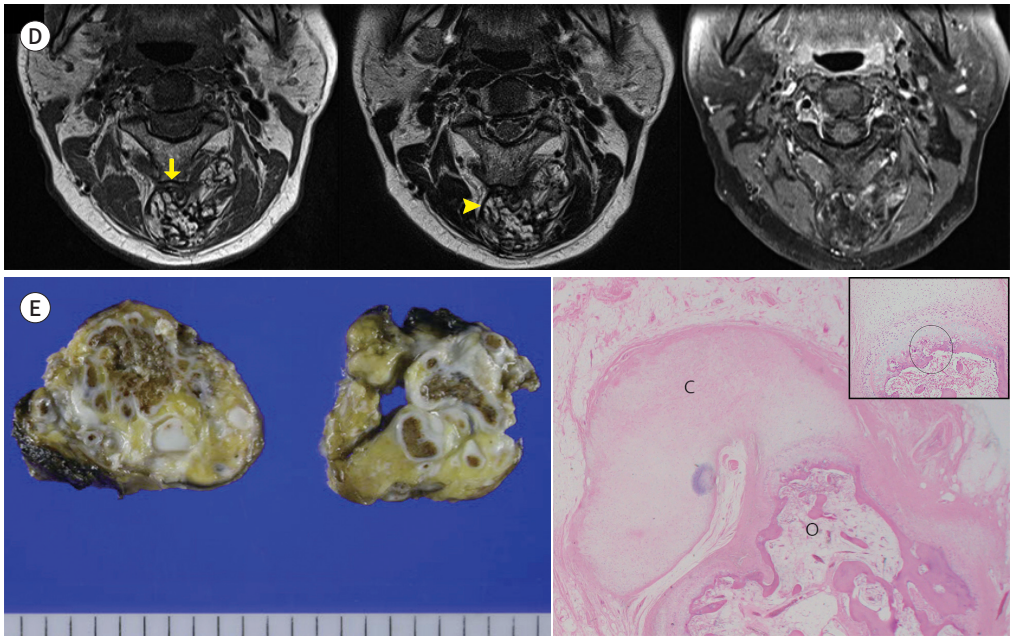
Fig. 1. Extraskeletal osteochondroma in the posterior neck of a 66-year-old female.
D. Axial T1WI, T2WI, and T1CE images reveal mild erosion at the C2 spinous process (arrow). The mass is located in the left posterior para-midline area, shifting the adjacent nuchal ligament to the right (arrowhead).
E. Photograph of the coronally sectioned gross specimen shows a solid-fibrotic mass with abundant adipose tissue, cartilage, and osteoid components. The low-power photomicrograph of the resected lesion (hematoxylin and eosin stain, × 12.5) shows an osteoid component (O) with a cartilage cap composed of mature hyaline cartilage, including an overlying fibrous perichondrium (C). Endochondral ossification with mature bone trabeculae beneath the cartilaginous cap is clearly visible in × 100 (Inlet).
T1CE = T1-weighted contrast-enhanced image, T1WI = T1-weighted image, T2WI = T2-weighted image



dochondral ossification with mature bone trabeculae located beneath the cartilaginous cap (Fig. 1E).

Our Institutional Review Board approved this case report and the requirement for informed consent was waived (IRB No. 2022GR0081).

DISCUSSION

Extraskeletal osteochondroma is a soft-tissue metaplasia characterized by the abnormal presence of mature cartilage and bone tissue. Typically, it arises in para-articular regions, including in digits of hands and feet (2). It is extremely rare to find it from outside synovial compartment, with only few sites including below 1st metatarsal bone shaft, within the thigh, and the nape of neck are reported (3-5). The pathogenesis of extraskeletal osteochondroma is unknown. It has been reported that mesenchymal cells in the connective tissue with progenitor capacity could give rise to cells of chondrogenic or osteogenic lineage that include chondroblasts, chondrocytes, osteoblasts, and osteocytes (6). Previous studies suggested repeated trauma may contribute to the development of this lesion (7). The standard treatment for extraskeletal osteochondroma is a local excision with preservation of the adjacent bone and soft tissue structures.

In radiologic examination, extraskeletal osteochondroma appears as a lobulated, round to

oval shaped mass localized in the soft tissue typically in para-articular regions without continuity with any bone. Calcification and/or ossification is usually present, which demonstrates corresponding densities or signal intensities in CT and MR (1, 2). In our case, a lobulated mass containing multiple amorphous and conglomerated ossified structures was present in posterior neck without bony continuation to cervical spine, which was compatible with the typical imaging finding of extraskeletal osteochondroma. Yet, the mass in our case was located far away from the joint, and demonstrated a significant amount of fatty background, which led us to false diagnosis as osteolipoma. Nakamura et al. (8) reported synovial osteochondroma, which is a variant of extraskeletal osteochondroma, arising in the spinal canal that also had fatty background. It has been suggested that extraskeletal osteochondroma may arise from metaplastic change of adipose tissue, which in turn may cause chondromatous and osseous transformation of the tissue (4). Thus, both cases support the previously suggested hypothesis, but the further studies should be conducted to fortify it.

Several other lesions may be considered in the differential diagnosis, including conventional osteochondroma, extraskeletal osteosarcoma and heterotopic ossification such as myositis ossificans, or any other ossified soft tissue tumors. Helpful radiologic findings for the correct diagnosis are the followings; conventional osteochondroma typically is an outgrowth of the long bones at the metaphysis that is continuous with the medullary cavity of the parent long bone (1). Extraskeletal osteosarcoma may also produce osteoid or cartilaginous matrix and appear as soft tissue mass with variable amounts of mineralization. It demonstrates heterogeneous enhancement in a degree depending on the amount of necrotic tissue present. The presence of hemorrhage is not rare (9). Myositis ossificans is a self-limiting benign inflammatory ossifying soft tissue mass usually within large muscles that typically occurs as a result of trauma. It has a zonal organization that is completed in 6–12 weeks, and it demonstrates a peripheral ossification with central lucency in mature stage (10).

In conclusion, we report a case of extraskeletal osteochondroma occurred in the posterior neck, far away from the joint without the involvement of joint components. Therefore, extraskeletal osteochondroma should be considered when a discrete, well-defined osseous mass without any direct continuity with the adjacent bone or joint is present in the soft tissues.

Author Contributions

Conceptualization, all authors; investigation, L.W.W.; supervision, H.S.; writing—original draft, L.W.W.; and writing—review & editing, H.S.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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중년 여성의 후경부에서 발생한 골격외 골연골종: 증례 보고

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골격외 골연골종은 연골종의 변종으로 일반적으로 족부와 수부의 관절 주위에서 발생한다. 이는 그 자체로도 희귀한 질환이지만 관절을 형성하는 구조물 외에서나 관절과 거리를 두고 발생하는 경우는 극히 드물다. 이 증례 보고는 66세 여성에게서 발생한 후경부의 골격외 골연골종의 사례이다. 저자들은 상기 질환의 전형적인 영상 소견 및 본 증례의 특이 소견을 문헌고찰과 함께 보고하고자 한다.

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