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Title: Paraarticular osteochondroma of the spine presenting as myelopathy: case report

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Abstract: Paraarticular osteochondroma is a rare osteocartilagenous tumor that arises in the soft tissue adjacent to a joint with no attachment to the bone. Although several case reports have been published on this tumor, spinal paraarticular osteochondroma has not been reported in the literature. We present a patient with a paraarticular osteochondroma arising in the spinal canal just medial to the facet joint that caused severe spinal cord compression. A 69-year-old man complained of paresthesia and muscle weakness in the lower extremities. Cervical magnetic resonance imaging and computed tomography revealed an intraspinal mass lesion at C7-Th1 with severe compression of the thecal sac, and concomitant spinal canal stenosis on C3-C7. Surgical en bloc resection of the calcified mass and C3-C6 laminoplasty were performed. The patient's symptom improved dramatically. Histological analysis showed that the lesion comprised trabecular bone and bone marrow, and was capped by hyaline cartilage with no connection to the bone. This is the first report of spinal paraarticular osteochondroma.

Suggested Reviewers:

1 **Paraarticular osteochondroma of the spine presenting as myelopathy:**
2 **case report**

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3 **Abstract**
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6 Paraarticular osteochondroma is a rare osteocartilagenous tumor that arises in the soft
7 tissue adjacent to a joint with no attachment to the bone. Although several case reports
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9 have been published on this tumor, spinal paraarticular osteochondroma has not been
10 reported in the literature. We present a patient with a paraarticular osteochondroma
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12 arising in the spinal canal just medial to the facet joint that caused severe spinal cord
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14 compression. A 69-year-old man complained of paresthesia and muscle weakness in the
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18 revealed an intraspinal mass lesion at C7–Th1 with severe compression of the thecal sac,
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20 and concomitant spinal canal stenosis on C3–C7. Surgical en bloc resection of the
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22 calcified mass and C3–C6 laminoplasty were performed. The patient’s symptom
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24 improved dramatically. Histological analysis showed that the lesion comprised
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26 trabecular bone and bone marrow, and was capped by hyaline cartilage with no
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28 connection to the bone. This is the first report of spinal paraarticular osteochondroma.
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3 **Introduction**
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6 Conventional osteochondroma is the most common type of benign bone tumor [1, 2]. It
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8 usually originates near the end of a long bone, and sometimes grows away from the
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10 joint. A osteochondroma is characterized by an osseous component capped by cartilage,
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12 and is connected to bone by a stalk [3]. Spinal osteochondroma comprises only 3% of
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14 solitary osteochondroma but may cause spinal cord compression [4-6]. A paraarticular
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16 or juxtaarticular osteochondroma is a rare osteocartilagenous tumor that arises in the
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18 soft tissue adjacent to a joint without attachment to bone [2, 3, 7]. Although several case
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20 reports have described paraarticular osteochondromas, all of the lesions reported were
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22 adjacent to the joints of the extremities, most of which involved the knee region [3, 8].
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24 To our knowledge, no case of spinal paraarticular osteochondromas has been reported.
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38 In this report, we present a patient with a paraarticular osteochondroma arising in the
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40 spinal canal just medial to the facet joint that caused severe spinal cord compression.
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47 **Case presentation**
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50 *History.* A 69-year-old man presented with paresthesia and muscle weakness in both
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52 lower extremities. He was referred to our hospital with a diagnosis of cervical
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54 spondylotic myelopathy. He occasionally stumbled when walking over slightly irregular
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3 surfaces, and had difficulty in walking up and down stairs. The symptoms were noticed
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6 one month before the consultation and then deteriorated rapidly. He had no history of
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9 osteochondroma in the extremities.
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12 *Examination.* His gait was spastic with a wide base, and tandem gait was unstable. His
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14 tendon reflexes were normal in the upper extremities and hyperactive in the lower legs
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18 with sustained ankle clonus. The Babinski sign was positive bilaterally. He had no
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22 clumsiness of the hands. Mild motor weakness was present in the left lower extremity.
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25 Superficial sensation was normal, but the sense of vibration was depressed in the lower
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28 extremities.
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31 *Radiography.* Plain radiographs of the cervical spine revealed spondylotic changes at
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35 C5–C7. Cervical MRI revealed an intraspinal mass lesion at C7–Th1 and severe
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38 compression of the thecal sac (Fig. 1). The lesion had a high intensity on both T₁– and
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41 T₂– weighted sequences. Spinal canal stenosis of C3–C7 was found. CT scanning
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44 showed that the lesion was located just medial to the right C7–Th1 facet joint and
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47 dorsolateral to the spinal cord. A thin calcified rim surrounded the central portion. (Fig.
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51 2). Spinal instability was not apparent.
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54 Surgical resection of the calcified mass and C3–C6 laminoplasty were planned
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57 because his neurological condition deteriorated rapidly.
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3 *Operation.* After laminectomy of C7–Th1, an epidural mass lesion was identified
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6 dorsolateral to the dura mater (Fig. 3). The lesion was connected to the ligamentum
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8 flavum but not to the facet joint or lamina. There was no adhesion between the dura and
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10 the lesion, which was resected easily. C3–C6 laminoplasty was also performed.
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15 *Histological findings.* Grossly, the lesion measured 15 × 9 × 6 mm, comprised multiple
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17 small nodules, and had a cartilaginous surface that was attached to the ligamentum
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19 flavum (Fig. 4). Histologically, the lesion comprised lamellar trabecular bone and bone
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21 marrow, and was capped by hyaline cartilage (Fig. 5a). Fibroblastic tissue was
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23 continuous with both bone and cartilage (Fig. 5b). Endochondral ossification intervened
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25 between the bone and cartilage. No feature suggestive of malignancy was found.
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34 *Postoperative Course.* The patient's muscle weakness and gait disturbance improved
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36 dramatically. The complete removal of the tumor and decompression of C3-7 were
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38 confirmed on 1 year postoperative MRI (Fig. 6)
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47 **Discussion**

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49 The concept of paraarticular osteochondroma was first introduced in 1958 by Jaffe,
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51 who used the synonymous terms paraarticular chondroma and intracapsular chondroma
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53 to describe osteochondral metaplasia occurring in the fibrous joint capsule or soft tissue
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3 adjacent to a joint [2, 9]. Milgram and Dunn were the first to use the term paraarticular
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6 osteochondroma and to differentiate the same lesion from synovial chondromatosis [7].
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10 About 35 cases were reported subsequently in the literature [3, 8, 10]. The knee is
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12 the most frequent site (76%), followed by the foot (19%) and ankle (5%) [3, 8]. Our
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14 report is the first on paraarticular osteochondroma arising in the spinal canal just medial
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17 to the articular facet joint.
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22 The diagnosis of this tumor is based on the association of radiological and histological
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24 features. Reith et al. established the following diagnostic criteria for paraarticular
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26 osteochondroma: (1) The lesion presented as a single, dominant mass, both
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28 radiographically and grossly. (2) The mass consisted histologically of both bone and
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30 cartilage, organized in a manner similar to conventional osteochondromas. (3) The
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32 lesion was not intra-articular, that is, it did not arise within the synovium itself [3]. The
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35 current case met these criteria.
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44 The pathological findings of paraarticular osteochondroma show certain characteristics
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46 [2, 3, 7, 8]. The lesion is a well circumscribed, mineralized mass without any direct
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48 continuity with the adjacent bone. Grossly, it comprises multiple osteochondral nodules.
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51 Histologically, the configuration of each nodule is similar to a conventional
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54 osteochondroma. The peripheral portion of each nodule contains cartilaginous areas,
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3 whereas the central portion contains lamellar, trabecular bone. Endochondral
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6 ossification is evident at the interface between the cartilage and bone. The histological
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9 features in the current case were also compatible with the diagnosis.

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12 The differential diagnosis of a mineralized, juxta-facet mass lesion in the spinal canal
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14 should include a spinal juxtafacet cyst, spinal osteochondroma, tumoral calcinosis,
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The differential diagnosis of a mineralized, juxta-facet mass lesion in the spinal canal should include a spinal juxtafacet cyst, spinal osteochondroma, tumoral calcinosis, synovial chondromatosis, and extraskeletal chondrosarcoma. A spinal juxtafacet cyst usually appears as an intraspinal extradural mass arising from the facet joint with degenerative changes. Although rare in the cervical spine, it preferentially involves the cervico-thoracic junction and often presents as myelopathy [11, 12]. Calcification of the cyst may also be present, but MRI reveals a hypointensity on T1-weighted images and a fluid-filled cystic lesion on T2 sequences [11, 13]. Spinal osteochondroma is characterized by an osseous stalk that is continuous either with the vertebral body, lamina, or pedicle [4-6]. Tumoral calcinosis is a periarticular tumor-like calcified mass that often occurs adjacent to the large joints of the extremities: it arises rarely in the spinal canal, and histologically lacks a cartilage cap [14, 15]. Paraarticular osteochondromas sometimes grow very large and show histological features suggestive of malignancy, including cytological atypia and hypercellularity of the cartilaginous component [3]. Therefore, it is sometimes important to distinguish this tumor from

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3 extraskelatal chondrosarcoma. In the current patient, the tumor was much smaller than
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6 those reported in the literature and lacked histological features of a malignancy, possibly
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9 because the intraspinal extradural location and presentation of myelopathy led to early
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12 detection of the tumor.
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16 Paraarticular osteochondromas are highly amenable to gross total removal and,
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19 although reports of follow-up of such cases are limited, no recurrences have been
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22 reported.
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26 As with other bone tumors, clinical and radiographic findings and histopathological
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29 examination are essential for a correct diagnosis.
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3 **Conflict of interest**
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6 The authors declare that they have no conflict of interest.
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3 **Figure Legend**
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6 Fig. 1. Intraspinous mass lesion at C7–T1 with severe compression of the spinal cord was
7 seen in sagittal T1-weighted (a), sagittal T2-weighted (b), and coronal T2-weighted (c)
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9 MR images. Spinal canal stenosis on C3–C7 was found concomitantly.
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13 Fig. 2. CT scanning showing the calcified lesion located just medial to the right C7–T1
14 facet joint and dorsolateral to the spinal cord (arrow).
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19 Fig. 3. After laminectomy of C7–T1, an epidural mass lesion was identified dorsolateral
20 to the dura mater (arrow).
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27 Fig. 4. Gross specimen showing multiple small nodules and a cartilaginous surface
28 attached to the ligamentum flavum (arrow).
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33 Fig. 5. (a) Microscopic examination showing lamellar trabecular bone and bone marrow
34 capped by hyaline cartilage. (Hematoxylin and eosin stain)
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41 (b) The fibroblastic tissue was continuous with both the bone and cartilage (arrows).
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44 (Hematoxylin and eosin stain)
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47 Fig. 6. One year postoperative MR imaging. The complete removal of the tumor and
48 decompression of C3-7 were confirmed.
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Fig 1(a)



Fig 1(b)



Fig 1(c)



Fig 2



Fig 3

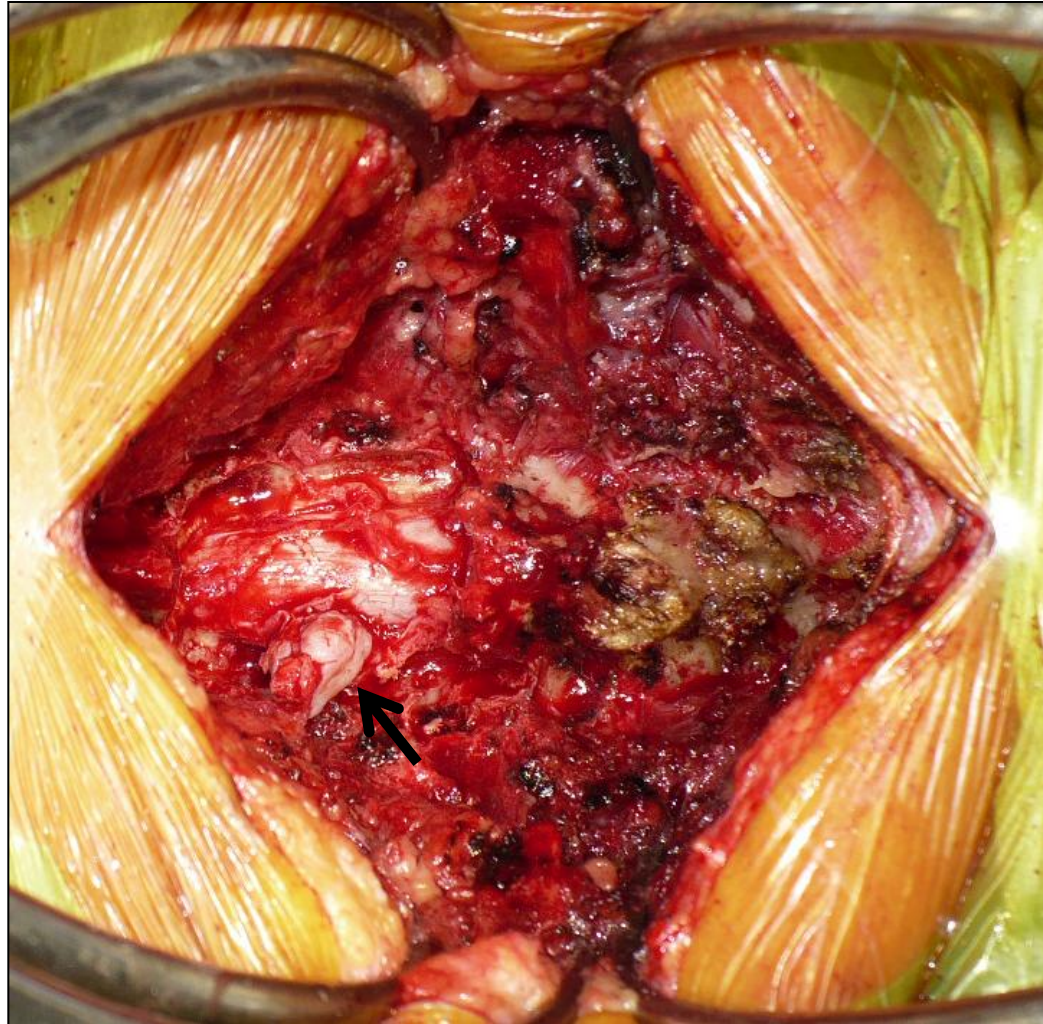


Fig 4



Fig 5(a)

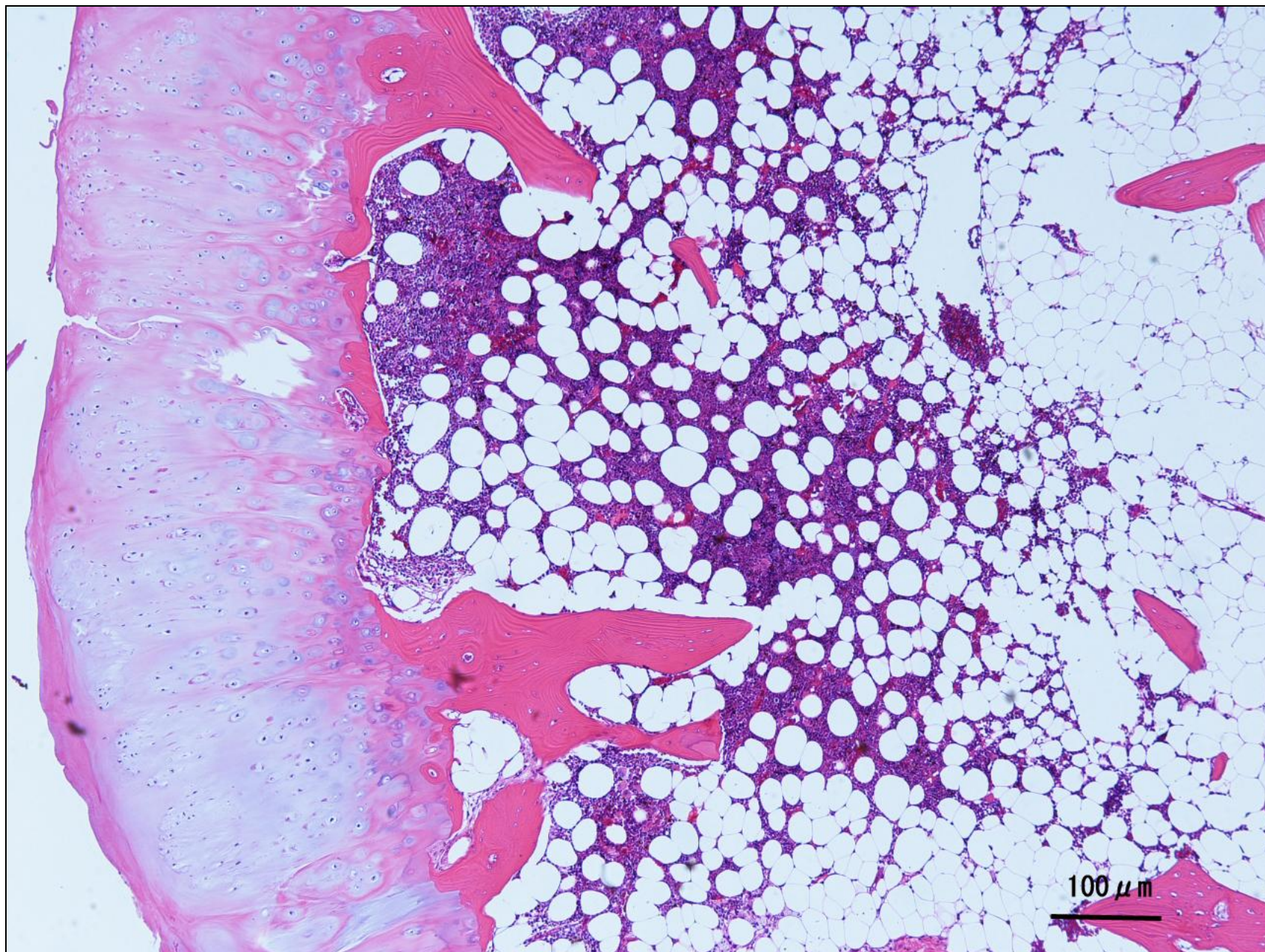


Fig 5(b)

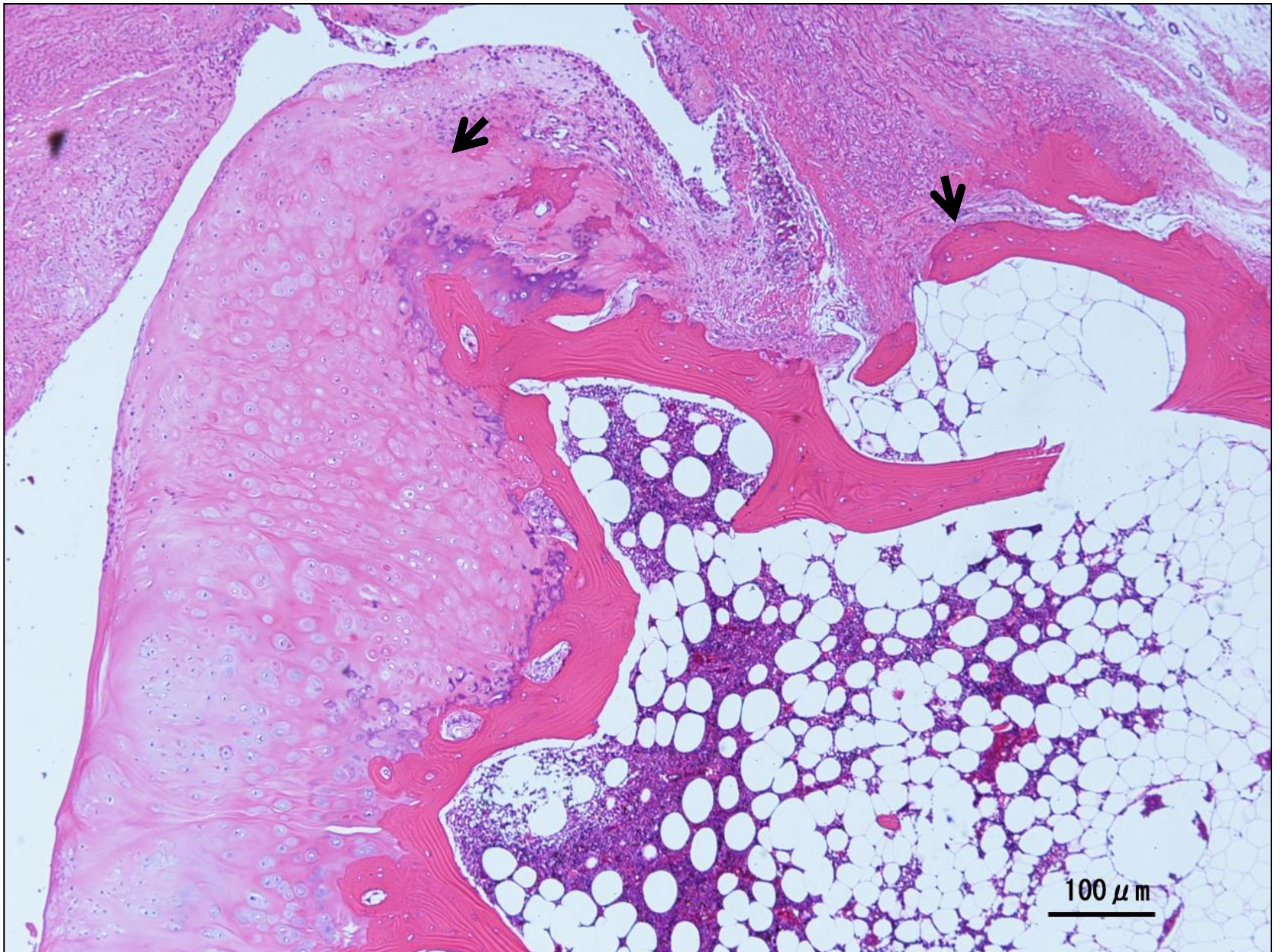


Fig 6

