

Editorial Manager(tm) for Skeletal Radiology
Manuscript Draft

Manuscript Number:

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

Article Type: Case Report

Keywords: Osteochondroma; Paraarticular osteochondroma; Soft tissue neoplasms; myelopathy

Corresponding Author: Takeshi Okamoto, M.D., Ph.D.

Corresponding Author's Institution: Kyoto University Graduate School of Medicine

First Author: Takeshi Okamoto, M.D., Ph.D.

Order of Authors: Takeshi Okamoto, M.D., Ph.D.; Masashi Neo, M.D., Ph.D.; Shunsuke Fujibayashi, M.D., Ph.D.; Mitsuru Takemoto, M.D., Ph.D.; Takashi Nakamura, M.D., Ph.D.

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**

3

4 **Takeshi Okamoto¹ MD, PhD, Masashi Neo¹ MD, PhD, Shunsuke Fujibayashi¹ MD,**
5 **PhD, Mitsuru Takemoto¹ MD, PhD, Takashi Nakamura¹ MD, PhD**

6

7 ¹Department of Orthopaedic Surgery, Graduate School of Medicine, Kyoto University,
8 Kyoto City, Japan

9

10 Corresponding Author:

11 Takeshi Okamoto

12 Department of Orthopaedic Surgery, Kyoto University Graduate School of Medicine,

13 54 Kawahara-cho, Shogoin, Sakyo-ku, Kyoto 606-8507, Japan

14 Tel: +81-75-751-3362

15 Fax: +81-75-751-8409

16 e-mail: okaken@kuhp.kyoto-u.ac.jp

17

18

1
2
3 **Abstract**
4
5

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
8
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
11
12 arising in the spinal canal just medial to the facet joint that caused severe spinal cord
13
14 compression. A 69-year-old man complained of paresthesia and muscle weakness in the
15
16 lower extremities. Cervical magnetic resonance imaging and computed tomography
17
18 revealed an intraspinal mass lesion at C7–Th1 with severe compression of the thecal sac,
19
20 and concomitant spinal canal stenosis on C3–C7. Surgical en bloc resection of the
21
22 calcified mass and C3–C6 laminoplasty were performed. The patient’s symptom
23
24 improved dramatically. Histological analysis showed that the lesion comprised
25
26 trabecular bone and bone marrow, and was capped by hyaline cartilage with no
27
28 connection to the bone. This is the first report of spinal paraarticular osteochondroma.
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60
61
62
63
64
65

1
2
3 **Introduction**
4

5
6 Conventional osteochondroma is the most common type of benign bone tumor [1, 2]. It
7
8 usually originates near the end of a long bone, and sometimes grows away from the
9
10 joint. A osteochondroma is characterized by an osseous component capped by cartilage,
11
12 and is connected to bone by a stalk [3]. Spinal osteochondroma comprises only 3% of
13
14 solitary osteochondroma but may cause spinal cord compression [4-6]. A paraarticular
15
16 or juxtaarticular osteochondroma is a rare osteocartilagenous tumor that arises in the
17
18 soft tissue adjacent to a joint without attachment to bone [2, 3, 7]. Although several case
19
20 reports have described paraarticular osteochondromas, all of the lesions reported were
21
22 adjacent to the joints of the extremities, most of which involved the knee region [3, 8].
23
24 To our knowledge, no case of spinal paraarticular osteochondromas has been reported.
25
26
27
28
29
30
31
32
33
34
35
36
37

38 In this report, we present a patient with a paraarticular osteochondroma arising in the
39
40 spinal canal just medial to the facet joint that caused severe spinal cord compression.
41
42
43
44
45
46

47 **Case presentation**
48

49
50 *History.* A 69-year-old man presented with paresthesia and muscle weakness in both
51
52 lower extremities. He was referred to our hospital with a diagnosis of cervical
53
54 spondylotic myelopathy. He occasionally stumbled when walking over slightly irregular
55
56
57
58
59
60

1
2
3 surfaces, and had difficulty in walking up and down stairs. The symptoms were noticed
4
5
6 one month before the consultation and then deteriorated rapidly. He had no history of
7
8
9 osteochondroma in the extremities.
10

11
12 *Examination.* His gait was spastic with a wide base, and tandem gait was unstable. His
13
14 tendon reflexes were normal in the upper extremities and hyperactive in the lower legs
15
16
17 with sustained ankle clonus. The Babinski sign was positive bilaterally. He had no
18
19
20
21
22 clumsiness of the hands. Mild motor weakness was present in the left lower extremity.
23
24
25 Superficial sensation was normal, but the sense of vibration was depressed in the lower
26
27
28
29 extremities.
30

31
32 *Radiography.* Plain radiographs of the cervical spine revealed spondylotic changes at
33
34
35 C5–C7. Cervical MRI revealed an intraspinal mass lesion at C7–Th1 and severe
36
37
38 compression of the thecal sac (Fig. 1). The lesion had a high intensity on both T₁– and
39
40
41 T₂– weighted sequences. Spinal canal stenosis of C3–C7 was found. CT scanning
42
43
44 showed that the lesion was located just medial to the right C7–Th1 facet joint and
45
46
47 dorsolateral to the spinal cord. A thin calcified rim surrounded the central portion. (Fig.
48
49
50
51 2). Spinal instability was not apparent.
52

53
54 Surgical resection of the calcified mass and C3–C6 laminoplasty were planned
55
56
57 because his neurological condition deteriorated rapidly.
58
59
60

1
2
3 *Operation.* After laminectomy of C7–Th1, an epidural mass lesion was identified
4
5
6 dorsolateral to the dura mater (Fig. 3). The lesion was connected to the ligamentum
7
8 flavum but not to the facet joint or lamina. There was no adhesion between the dura and
9
10 the lesion, which was resected easily. C3–C6 laminoplasty was also performed.
11
12
13

14
15 *Histological findings.* Grossly, the lesion measured 15 × 9 × 6 mm, comprised multiple
16
17 small nodules, and had a cartilaginous surface that was attached to the ligamentum
18
19 flavum (Fig. 4). Histologically, the lesion comprised lamellar trabecular bone and bone
20
21 marrow, and was capped by hyaline cartilage (Fig. 5a). Fibroblastic tissue was
22
23 continuous with both bone and cartilage (Fig. 5b). Endochondral ossification intervened
24
25 between the bone and cartilage. No feature suggestive of malignancy was found.
26
27
28
29
30
31
32

33
34 *Postoperative Course.* The patient’s muscle weakness and gait disturbance improved
35
36 dramatically. The complete removal of the tumor and decompression of C3-7 were
37
38 confirmed on 1 year postoperative MRI (Fig. 6)
39
40
41
42
43
44
45
46

47 **Discussion**

48
49 The concept of paraarticular osteochondroma was first introduced in 1958 by Jaffe,
50
51 who used the synonymous terms paraarticular chondroma and intracapsular chondroma
52
53 to describe osteochondral metaplasia occurring in the fibrous joint capsule or soft tissue
54
55
56
57
58
59
60
61
62
63
64
65

1
2
3 adjacent to a joint [2, 9]. Milgram and Dunn were the first to use the term paraarticular
4
5
6 osteochondroma and to differentiate the same lesion from synovial chondromatosis [7].
7
8

9
10 About 35 cases were reported subsequently in the literature [3, 8, 10]. The knee is
11
12 the most frequent site (76%), followed by the foot (19%) and ankle (5%) [3, 8]. Our
13
14 report is the first on paraarticular osteochondroma arising in the spinal canal just medial
15
16 to the articular facet joint.
17
18
19
20
21

22 The diagnosis of this tumor is based on the association of radiological and histological
23
24 features. Reith et al. established the following diagnostic criteria for paraarticular
25
26 osteochondroma: (1) The lesion presented as a single, dominant mass, both
27
28 radiographically and grossly. (2) The mass consisted histologically of both bone and
29
30 cartilage, organized in a manner similar to conventional osteochondromas. (3) The
31
32 lesion was not intra-articular, that is, it did not arise within the synovium itself [3]. The
33
34 current case met these criteria.
35
36
37
38
39
40
41
42
43

44 The pathological findings of paraarticular osteochondroma show certain characteristics
45
46 [2, 3, 7, 8]. The lesion is a well circumscribed, mineralized mass without any direct
47
48 continuity with the adjacent bone. Grossly, it comprises multiple osteochondral nodules.
49
50
51 Histologically, the configuration of each nodule is similar to a conventional
52
53 osteochondroma. The peripheral portion of each nodule contains cartilaginous areas,
54
55
56
57
58
59
60

1
2
3 whereas the central portion contains lamellar, trabecular bone. Endochondral
4
5
6 ossification is evident at the interface between the cartilage and bone. The histological
7
8
9 features in the current case were also compatible with the diagnosis.

10
11
12 The differential diagnosis of a mineralized, juxta-facet mass lesion in the spinal canal
13
14 should include a spinal juxtafacet cyst, spinal osteochondroma, tumoral calcinosis,
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60
61
62
63
64
65
66
67
68
69
70
71
72
73
74
75
76
77
78
79
80
81
82
83
84
85
86
87
88
89
90
91
92
93
94
95
96
97
98
99
100
101
102
103
104
105
106
107
108
109
110
111
112
113
114
115
116
117
118
119
120
121
122
123
124
125
126
127
128
129
130
131
132
133
134
135
136
137
138
139
140
141
142
143
144
145
146
147
148
149
150
151
152
153
154
155
156
157
158
159
160
161
162
163
164
165
166
167
168
169
170
171
172
173
174
175
176
177
178
179
180
181
182
183
184
185
186
187
188
189
190
191
192
193
194
195
196
197
198
199
200
201
202
203
204
205
206
207
208
209
210
211
212
213
214
215
216
217
218
219
220
221
222
223
224
225
226
227
228
229
230
231
232
233
234
235
236
237
238
239
240
241
242
243
244
245
246
247
248
249
250
251
252
253
254
255
256
257
258
259
260
261
262
263
264
265
266
267
268
269
270
271
272
273
274
275
276
277
278
279
280
281
282
283
284
285
286
287
288
289
290
291
292
293
294
295
296
297
298
299
300
301
302
303
304
305
306
307
308
309
310
311
312
313
314
315
316
317
318
319
320
321
322
323
324
325
326
327
328
329
330
331
332
333
334
335
336
337
338
339
340
341
342
343
344
345
346
347
348
349
350
351
352
353
354
355
356
357
358
359
360
361
362
363
364
365
366
367
368
369
370
371
372
373
374
375
376
377
378
379
380
381
382
383
384
385
386
387
388
389
390
391
392
393
394
395
396
397
398
399
400
401
402
403
404
405
406
407
408
409
410
411
412
413
414
415
416
417
418
419
420
421
422
423
424
425
426
427
428
429
430
431
432
433
434
435
436
437
438
439
440
441
442
443
444
445
446
447
448
449
450
451
452
453
454
455
456
457
458
459
460
461
462
463
464
465
466
467
468
469
470
471
472
473
474
475
476
477
478
479
480
481
482
483
484
485
486
487
488
489
490
491
492
493
494
495
496
497
498
499
500
501
502
503
504
505
506
507
508
509
510
511
512
513
514
515
516
517
518
519
520
521
522
523
524
525
526
527
528
529
530
531
532
533
534
535
536
537
538
539
540
541
542
543
544
545
546
547
548
549
550
551
552
553
554
555
556
557
558
559
560
561
562
563
564
565
566
567
568
569
570
571
572
573
574
575
576
577
578
579
580
581
582
583
584
585
586
587
588
589
590
591
592
593
594
595
596
597
598
599
600
601
602
603
604
605
606
607
608
609
610
611
612
613
614
615
616
617
618
619
620
621
622
623
624
625
626
627
628
629
630
631
632
633
634
635
636
637
638
639
640
641
642
643
644
645
646
647
648
649
650
651
652
653
654
655
656
657
658
659
660
661
662
663
664
665
666
667
668
669
670
671
672
673
674
675
676
677
678
679
680
681
682
683
684
685
686
687
688
689
690
691
692
693
694
695
696
697
698
699
700
701
702
703
704
705
706
707
708
709
710
711
712
713
714
715
716
717
718
719
720
721
722
723
724
725
726
727
728
729
730
731
732
733
734
735
736
737
738
739
740
741
742
743
744
745
746
747
748
749
750
751
752
753
754
755
756
757
758
759
760
761
762
763
764
765
766
767
768
769
770
771
772
773
774
775
776
777
778
779
780
781
782
783
784
785
786
787
788
789
790
791
792
793
794
795
796
797
798
799
800
801
802
803
804
805
806
807
808
809
810
811
812
813
814
815
816
817
818
819
820
821
822
823
824
825
826
827
828
829
830
831
832
833
834
835
836
837
838
839
840
841
842
843
844
845
846
847
848
849
850
851
852
853
854
855
856
857
858
859
860
861
862
863
864
865
866
867
868
869
870
871
872
873
874
875
876
877
878
879
880
881
882
883
884
885
886
887
888
889
890
891
892
893
894
895
896
897
898
899
900
901
902
903
904
905
906
907
908
909
910
911
912
913
914
915
916
917
918
919
920
921
922
923
924
925
926
927
928
929
930
931
932
933
934
935
936
937
938
939
940
941
942
943
944
945
946
947
948
949
950
951
952
953
954
955
956
957
958
959
960
961
962
963
964
965
966
967
968
969
970
971
972
973
974
975
976
977
978
979
980
981
982
983
984
985
986
987
988
989
990
991
992
993
994
995
996
997
998
999
1000

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

1
2
3 extraskelatal chondrosarcoma. In the current patient, the tumor was much smaller than
4
5
6 those reported in the literature and lacked histological features of a malignancy, possibly
7
8
9 because the intraspinal extradural location and presentation of myelopathy led to early
10
11
12 detection of the tumor.
13

14
15
16 Paraarticular osteochondromas are highly amenable to gross total removal and,
17
18
19 although reports of follow-up of such cases are limited, no recurrences have been
20
21
22 reported.
23

24
25
26 As with other bone tumors, clinical and radiographic findings and histopathological
27
28
29 examination are essential for a correct diagnosis.
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60
61
62
63
64
65

1
2
3 **Conflict of interest**
4
5

6 The authors declare that they have no conflict of interest.
7
8
9

10
11
12 **References**
13

- 14
15
16 1. Unni KK, Inwards CY. Osteochondroma. In: Dahlin's Bone Tumors. Philadelphia:
17
18 Lippincott Williams & Wilkins, 2010: 9–21
19
20
21
22 2. Maheshwari AV, Muro-Cacho CA, Pitcher JD. Extraskeletal para-articular
23
24 osteochondroma of the posterior knee. Case report. J Knee Surg 2009; 22: 30–33
25
26
27
28 3. Reith JD, Bauer TW, Joyce MJ. Paraarticular osteochondroma of the knee. Report of
29
30 2 cases and review of the literature. Clin Orthop Relat Res 1997; 334: 225–232
31
32
33
34
35 4. Lotfinia I, Vahedi P, Tubbs RS, et al. Neurological manifestations, imaging
36
37 characteristics, and surgical outcome of intraspinal osteochondroma. J Neurosurg
38
39 Spine 2010; 12: 474–489
40
41
42
43
44 5. Roach JW, Klatt JW, Faulkner ND. Involvement of the spine in patients with
45
46 multiple hereditary exostoses. J Bone Joint Surg Am 2009; 91:1942–1948
47
48
49
50
51 6. Yagi M, Ninomiya K, Kihara M, et al. Symptomatic osteochondroma of the spine in
52
53 elderly patients. Reports of 3 cases. J Neurosurg Spine 2009; 11: 64–70
54
55
56
57 7. Milgram JW, Dunn EJ. Para-articular chondromas and osteochondromas: A report of
58
59
60

- 1
2
3 three cases. *Clin Orthop Relat Res* 1980; 148: 147–151
4
5
6
7 8. Ratcliff JR, Naqvi A, de la Roza G, et al. Soft tissue osteochondroma; case report
8
9 and immunohistochemistry for parathyroid hormone-related protein. *Ann Diagn*
10
11
12
13 *Pathol* 2006; 10(4): 222–229
14
15
16 9. Jaffe HL. *Tumors and timorous conditions of the bones and joints*. Philadelphia: Lea
17
18
19 and Febiger 1958: 558–567
20
21
22 10. Ozturan KE, Yucel U, Cakici H, et al. Patellar tendinopathy caused by a
23
24
25 para-articular/extraskeletal osteochondroma in the lateral infrapatellar region of the
26
27
28 knee: a case report. *Cases J* 2009; 2: 1–4
29
30
31 11. Song JK, Musleh W, Christie SD, et al. Cervical juxtafacet cysts: case report and
32
33
34
35 literature review. *Spine J* 2006; 6: 279–281
36
37
38 12. Miwa M, Doita M, Takayama H, et al. An expanding cervical synovial cyst causing
39
40
41 acute cervical radiculopathy. *J Spinal Disord Tech* 2004; 17: 331–333
42
43
44 13. Nijensohn E, Russell EJ, Milan M, et al. Calcified synovial cyst of the cervical
45
46
47 spine: CT and MR evaluation. *J Comput Assist Tomogr* 1990; 14(3): 473–476
48
49
50 14. Miyakoshi N, Shimada Y, Kasukawa Y, et al. Progressive myelopathy due to
51
52
53 idiopathic intraspinal tumoral calcinosis of the cervical spine. *J Neurosurg Spine*
54
55
56
57 2007; 7: 362–365
58
59
60
61
62
63
64
65

1
2
3 15. Flores J, Gallego JM, Bujan A, et al. Spinal cord compression due to tumoral
4
5
6 idiopathic calcinosis. Spinal Cord 2003; 41: 413–416
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60
61
62
63
64
65

1
2
3 **Figure Legend**
4
5

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)
8 MR images. Spinal canal stenosis on C3–C7 was found concomitantly.
9

10
11
12
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).
15

16
17
18
19 Fig. 3. After laminectomy of C7–T1, an epidural mass lesion was identified dorsolateral
20 to the dura mater (arrow).
21

22
23
24
25 Fig. 4. Gross specimen showing multiple small nodules and a cartilaginous surface
26 attached to the ligamentum flavum (arrow).
27

28
29
30
31 Fig. 5. (a) Microscopic examination showing lamellar trabecular bone and bone marrow
32 capped by hyaline cartilage. (Hematoxylin and eosin stain)
33

34
35
36
37 (b) The fibroblastic tissue was continuous with both the bone and cartilage (arrows).
38
39
40
41
42
43
44
45 (Hematoxylin and eosin stain)
46

47
48 Fig. 6. One year postoperative MR imaging. The complete removal of the tumor and
49 decompression of C3-7 were confirmed.
50
51
52
53
54
55
56
57
58
59
60

Fig 1(a)



Fig 1(b)



Fig 1(c)



Fig 2



Fig 3

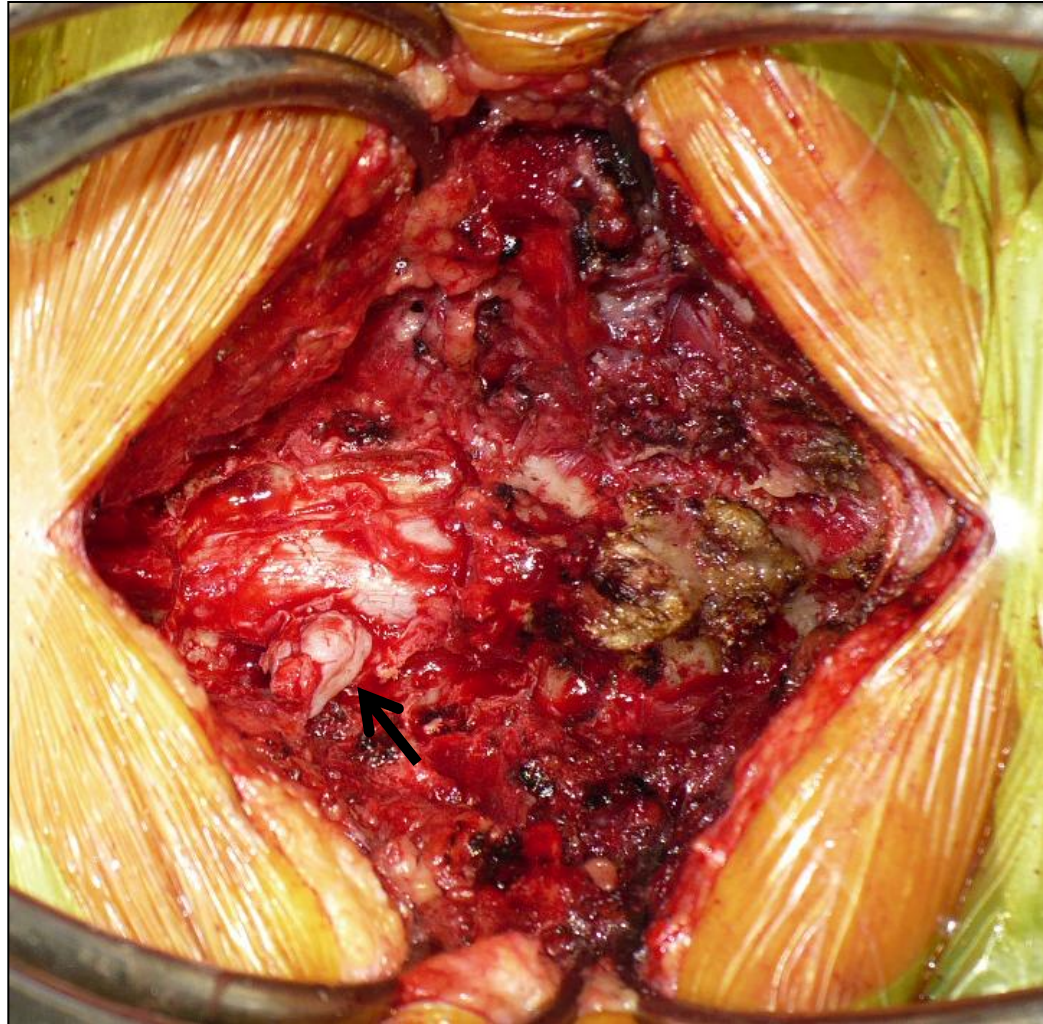


Fig 4



Fig 5(a)

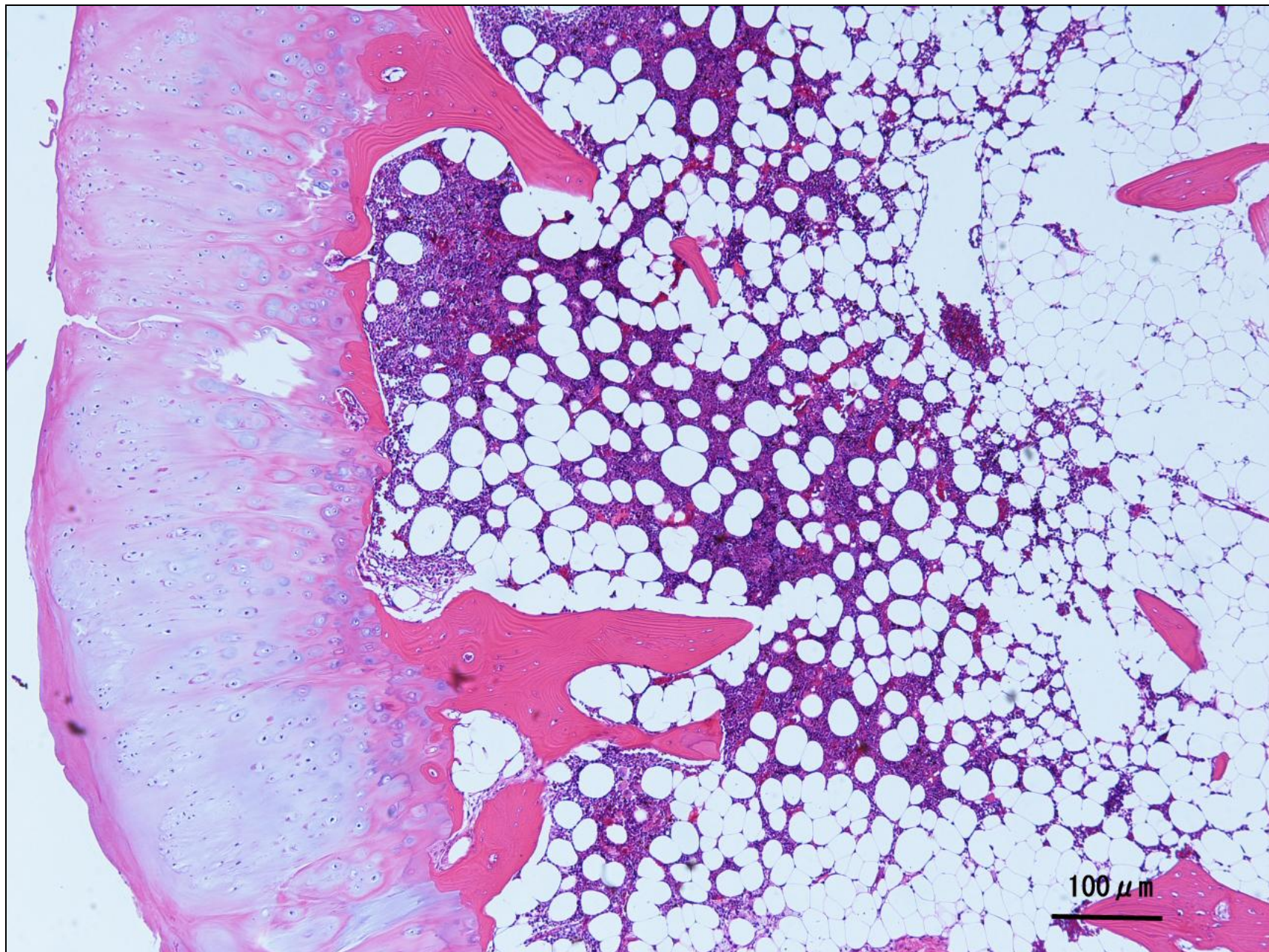


Fig 5(b)

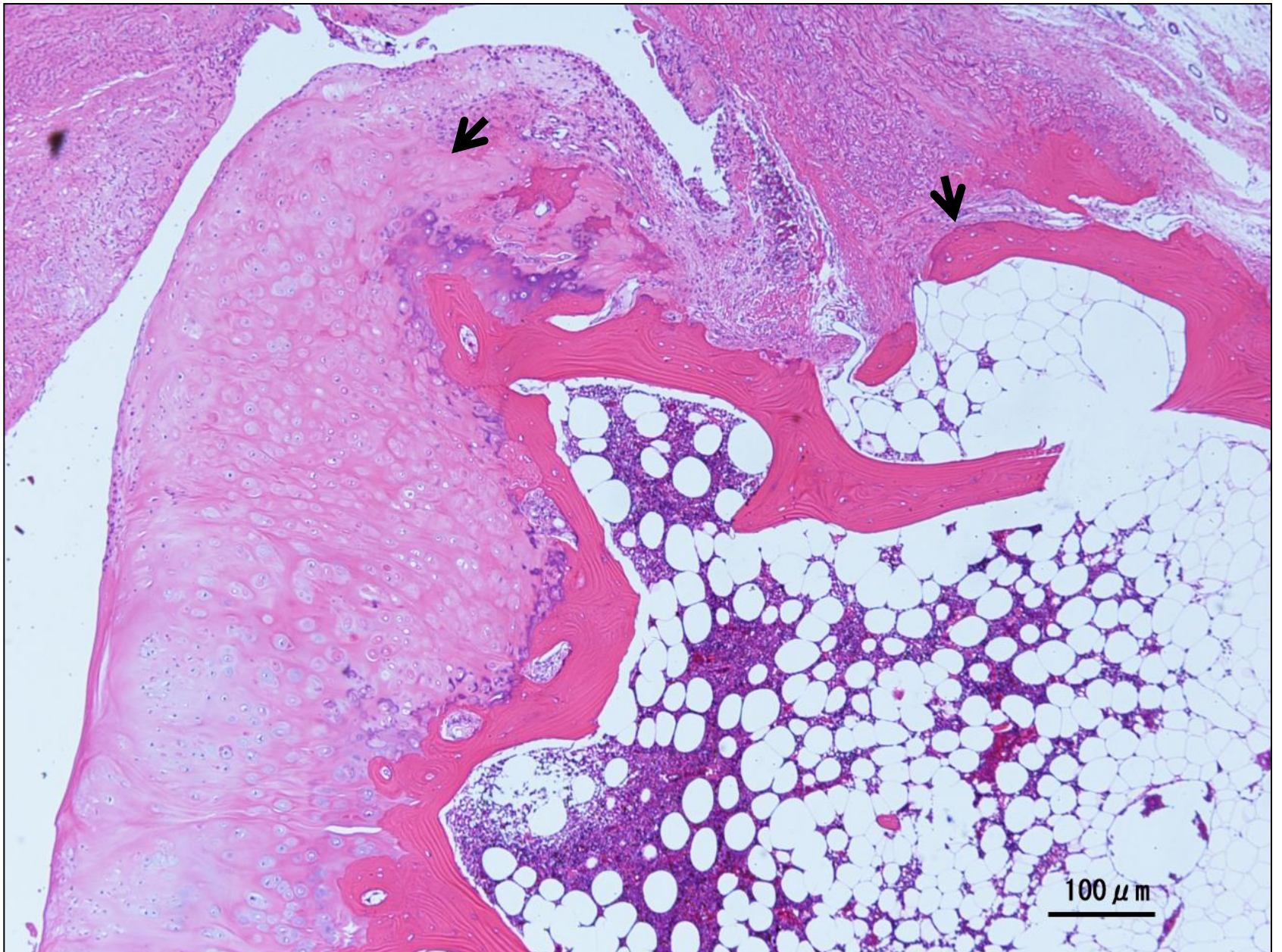


Fig 6

