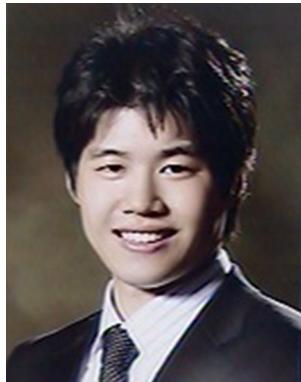


## Cervical subtotal en-bloc spondylectomy of C6 mesenchymal chondrosarcoma

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### Abstract



**Introduction** We present a case of C6 mesenchymal chondrosarcoma and discuss safe posterior to anterior approach subtotal en-bloc spondylectomy.

**Materials and methods** A 29-year-old male consulted for our department with severe posterior neck pain doing exercise. CT scan demonstrated a primary osteolytic lesion on C6 left transverse foramen and MRI demonstrated the

tumor involved C6 vertebra from layers B, C and F sectors 4–6 encasing left vertebral artery. Preoperatively neurointerventional radiology service occluded the left vertebral artery and tumor feeding artery using coil embolization. Posterior approach consist of C5–C7 laminectomy, left sided C6 and C7 nerve root sacrifice, posterior disc removal and release of C5–6–7 and posterior reconstruction. Then, position was changed to supine, and the anterior approach was followed as C5–6, C6–7 discectomy, left vertebral artery ligation and cut, longus colli resection and C6 subtotal spondylectomy with en-bloc resection of mass, mesh cage insertion and C5–C7 anterior plate fixation. During operation, frozen biopsy was performed on 8 areas (longus colli, lateral margin, anteroinferior margin, posterior margin, posterosuperior margin, C5 transverse foramen, posteroinferior margin, inferior margin) after wide resection. Tumor free margin was confirmed.

**Results** After operation, he complained of tingling sensation of left thumb and forearm medial side, and elbow extensor motor grade was checked to 4/5 postoperatively. In the followed-up radiograph, the tumor was completely removed, and the instability of joint was not seen. As a result of observing follow-up CT at a year after the surgery, recurrence findings have not been shown up to now, and the progression of neurologic symptoms has not been shown either.

**Conclusion** Based on the Grand Round case and relevant literature, we discuss the case of mesenchymal chondrosarcoma occurring from the C6 cervical spine treated with cervical subtotal en-bloc spondylectomy. Successful en-bloc resection of the tumor was achieved using posterior to anterior approach.

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**Keywords** Cervical spine tumor · Mesenchymal chondrosarcoma · Subtotal en-bloc spondylectomy

## Case presentation

A 29-year-old male consulted for our department with severe posterior neck pain during exercise. There was no motor weakness or numbness on physical examination. He had undergone medical and physical therapy over the 2 months, but pain was not resolved. He was investigated by way of plain radiograph, CT scan and MRI scan. Bony mass was confirmed at CT and MRI. A biopsy was performed under a C-arm guide and the reported diagnosis was mesenchymal chondrosarcoma. Positron emission tomography-CT (PET-CT) demonstrated no distal metastasis, therefore planned C6 subtotal en-bloc spondylectomy with sacrifice of left vertebral artery, C6 root and C7 root through posterior and anterior surgical approach.

## Diagnostic imaging

Plain radiographs showed nonspecific feature, but computed tomography (CT) scan demonstrated osteolytic lesion on C6 left lateral mass and transverse process (Fig. 1). Magnetic resonance imaging (MRI) demonstrated a mass surrounding vertebral foramen on C6 vertebral body and tumor involved layer B, C and F, sectors 4–6 and there was no cord compression [1] (Fig. 2a, b).

## Historical review

Chondrosarcoma is the second most common sarcoma of the skeleton after osteosarcoma and the most frequent sites of chondrosarcoma are the pelvis [2]. Chondrosarcoma of



**Fig. 1** C6 left transverse foramen osteolytic lesion is observed in CT axial cut

the spine is not occurred commonly (4–10 %) compared to that in the long bones, and mesenchymal chondrosarcoma is extremely rare malignant tumor [3]. Mesenchymal chondrosarcoma occur in both the bone and soft tissue, and several reports of this tumor occurring in the maxilla, mandible, CNS system [4]. Main prognostic factor of chondrosarcoma include histologic feature, patient age, feasibility of performing en-bloc resection with appropriate oncologic margins [5].

In spine, owing to its proximity to neurovascular structure, to perform en-bloc resection is difficult for the surgeon and has poor prognosis. Therefore, whether resection can be conducted including appropriate oncologic margin may reduce local and metastatic recurrence and be an important point related to the fall of mortality, which is reported in a number of clinical data [6, 7].

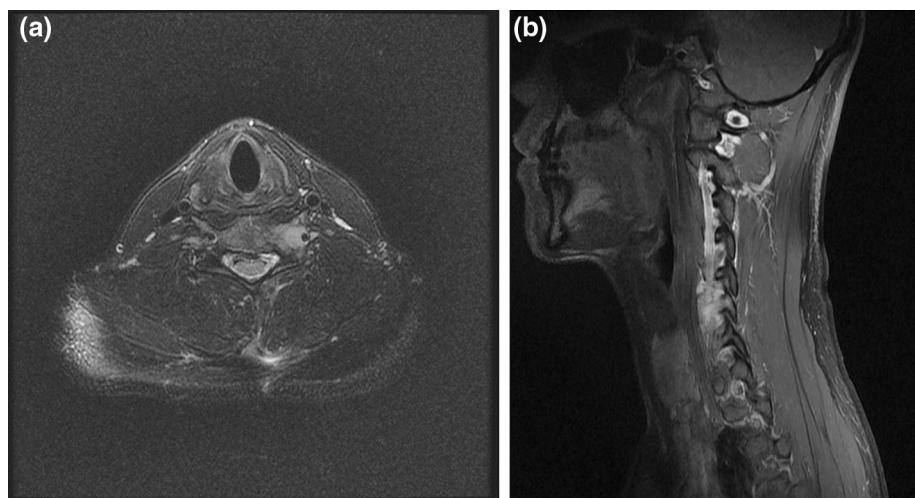
Total en-bloc spondylectomy is reported to be one of the most effective surgical techniques for spinal tumors and satisfactory results after total en-bloc spondylectomy for osteosarcoma of the spine was reported [8–10]. However, several factors complicate performing this technique in the cervical spine unlike in thoracolumbar spine and sacrum, including the proximity of the vertebral arteries, the complex bony structure, and the importance of the cervical nerve roots [11].

## Rationale for treatment and evidence-based literature

This is cervical mesenchymal chondrosarcoma, which is a very rare case report, and it is applicable to IIB of Enneking stage [12]. In cervical spine, no en-bloc resection has been performed in chondrosarcoma of Enneking stage IIB. Xinghai et al. [13] conducted extracapsular piecemeal resection in four cases of IIB, and en-bloc resection in three cases. In addition to these, there have been five case reports of en-bloc resection with chordoma diagnosed in the cervical vertebra [11, 14]. Although it is reported in a lot of literature that en-bloc resection can be indicated if there is no sign of disseminated disease (lesions Enneking grade IA, IB, IIA, or IIB), en-bloc resection in spine may be difficult to be performed because of complex anatomy and neurovascular structure, and it would be more so in cervical spine. Therefore, ideal en-bloc resection may not be conducted with sacrifice of spinal cord or nerve root. For ideal en-bloc resection in cervical spine region, spinal cord ligation has been reported, but it needs more consideration regarding subsequent severe neurologic deficit patients may receive [15].

There have been no established theories on the indication of total en-bloc resection in cervical spine. En-bloc excision in cervical spine is technically demanding, and it

**Fig. 2** In T2 MRI axial cut (a) and sagittal cut (b), mass lesion which encircles C6 left vertebral artery, and invades C6 vertebral body and even lateral mass was identified



has been considered impossible because of vertebral artery and cervical nerve root, but case reports on it are gradually reported [16]. Boriani et al. [17] performed en-bloc resection in four cases limited to posterior element ranged from WBB sector 10 to 3. Leitner et al. [14] stated that if a case is limited to WBB sector 6.7, and it does not invade layer D or more in small tumor, resection may be possible with adequate tumor free margin, and it is adequate for low-grade malignancy such as aggressive benign tumor or chordoma. If tumor invaded both anterior column and posterior column, en-bloc resection would require very technically demanding, and especially in cervical spine, a few cases were rarely reported [18–20].

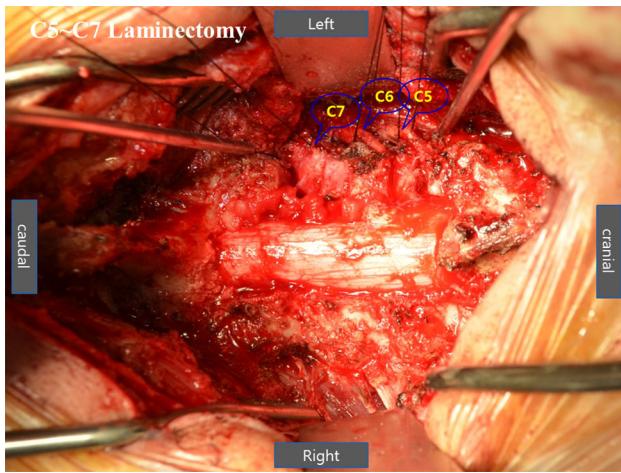
This case was layer B, C, F sectors 4–6 in WBB classification, posterior component and root sacrifice were conducted through posterior surgical approach. After that, spondylectomy was conducted, and vertebral artery was removed using anterior surgical approach. In a number of studies, it was first approached in posterior when conducted in combined approach, but its reason was not exactly described. It will be more advantageous to facilitate sacrifice of nerve root and vertebral artery and perform en-bloc resection with adequate margin by conducting posterior approach first being separated from spinal cord beforehand. Releasing posterior vertebral column between spinal cord and vertebral body after conducting total C6 laminectomy through posterior approach to secure margin, and then performing cervical spondylectomy through anterior approach will help reduce time and conduct the operation accurately. Subtotal en-bloc spondylectomy with adequate margin was successfully conducted except for cord region, but to confirm the therapeutic effectiveness, at least 5 years of follow-up observation will be needed. For chondrosarcoma, because adjuvant therapy such as chemotherapy or radiative therapy is not effective, the resection of tumor will be the

most effective therapy. If primary metastasis did not occur, the future plan should be established after understanding the risk of en-bloc resection in cervical spine mentioned above.

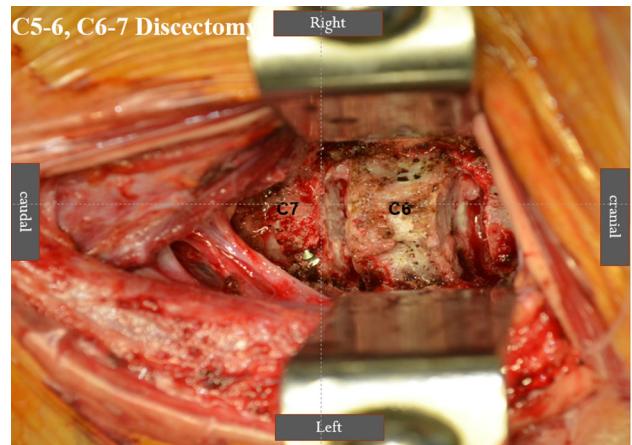
Proton beam therapy has been indicated adjuvantly for primary spinal tumor like chordoma and chondrosarcoma, which had advantage of sparing normal tissues from unnecessary exposure. In our case, the tumor was diagnosed as mesenchymal chondrosarcoma and it did not respond to radiative therapy completely, therefore we did not consider proton beam therapy.

As the major risk and complications related to en-bloc resection in cervical spine, excessive bleeding, accidental injury in spinal cord, nerve root, and main blood vessels, possibilities of contamination of tumor cells, severe dysphagia, aspiration pneumonia, post-operative cervical instability may occur [11]. To prevent excessive bleeding, it is needed to use fibrin glue in preoperative embolization and epidural vein, and meticulous blunt resection is needed [7, 21]. In this case preoperative left vertebra artery and tumor nutrient artery embolization procedure were performed, so the bleeding could be prevented in a certain degree by embolization procedure with about 800 cc of bleeding during the operation. In addition, by conducting combined anterior and posterior approach and fusion, spine was fixed strongly to prevent the cervical vertebral instability.

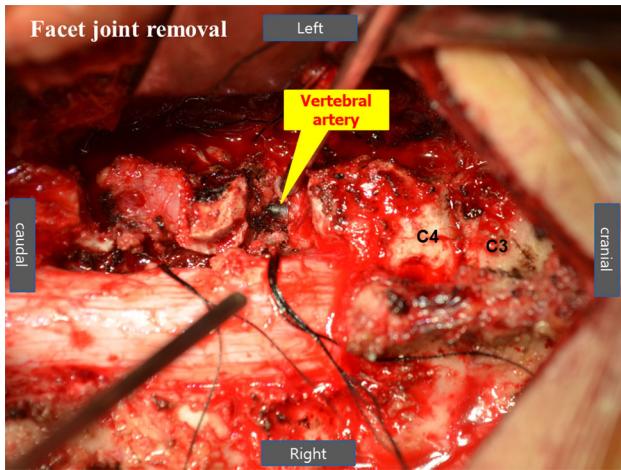
There are still controversies on clipping of vertebral artery, but many articles have been reported ligation of vertebral artery for removal of tumor in cervical vertebra [18, 19, 22]. Hoshino et al. [23] reported that unilateral vertebral artery did not affect due to brain stem, cerebellum, and ischemic injury of spinal cord after clipping. In this case, additional cerebrovascular and neurologic complications did not occur after performing preoperative left vertebra embolization.



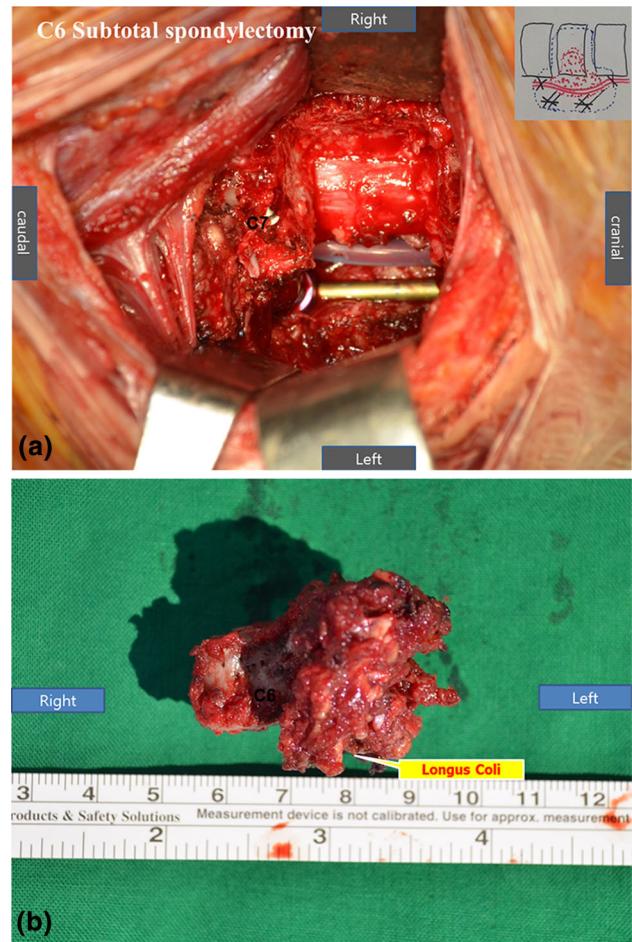
**Fig. 3** By performing C5–C7 laminectomy, cord and C7 nerve root were exposed, and C6, C7 nerve roots were tied



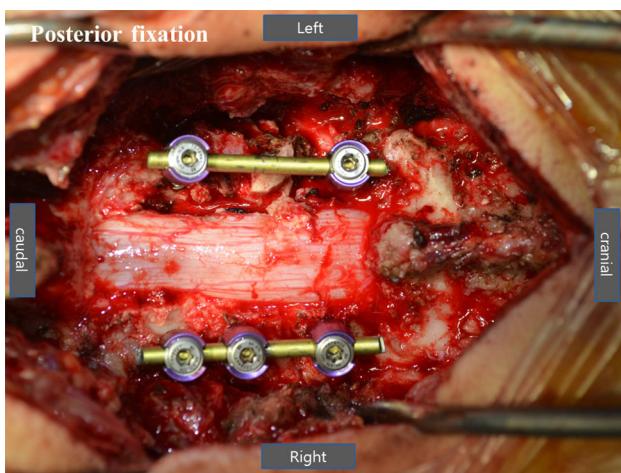
**Fig. 6** After exposing C5–C7, C5–6 and C6–7 discectomy was performed, and then joint of Luschka was removed



**Fig. 4** C5 inferior facet, C6 superior and inferior facet and C7 superior facet joint were removed, and Lt vertebral artery filled with thrombus could be identified



**Fig. 7** **a** After cutting in right C6 vertebral body in an oblique way, the tumor was removed by performing subtotal spondylectomy. **b** Part of longus colli is attached in C6 body taken in 0.5 cm and anterior site



**Fig. 5** Lateral mass screw was fixated at C5 and C6, and pedicle screw was fixated to C7, and then wound was sutured

## Procedure

Preoperatively neurointerventional radiology service occluded the left vertebral artery and tumor feeding artery using coil embolization to minimize bleeding from tumor.

### Anterior approach

For surgical technique, the patient was laid in prone position, and exposure was performed from C2 vertebra to T1 vertebra with posterior approach. Then, left lateral mass was partially resected, C5 root and C6 root were exposed, C5, C6, and C7 laminectomy was performed to expose spinal cord and C7 root, and ligation was conducted in C6 root and C7 root (Fig. 3). C5 left inferior facet, C6 left superoinferior facet, and C7 superior facet were removed,



**Fig. 8** After removing tumor, wide resection was extensively performed in eight regions including longus colli, lateral margin, anteroinferior margin, posterior margin, posterosuperior margin, C5 transverse foramen, posteroinferior margin, and inferior margin, and then cage was inserted

**Fig. 9 a, b** C-spine AP/lateral view taken after surgery

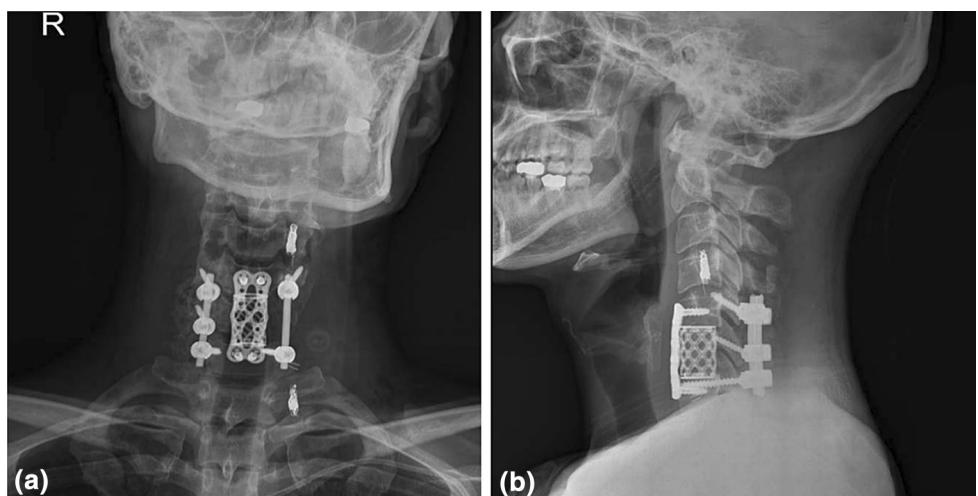
and the left vertebral artery filled with thrombus was identified after coiling when removing C6 left superior facet (Fig. 4). After that, joint was removed, ligated C6 root and C7 root were cut, and C5–6 and C6–7 discectomy and C6 body posterior release were performed. Then, after screws were fixed in C5 both lateral mass, C6 right lateral mass, and C7 both pedicle, rod was connected and fixed, and surgical wound suture was performed (Fig. 5).

### Posterior approach

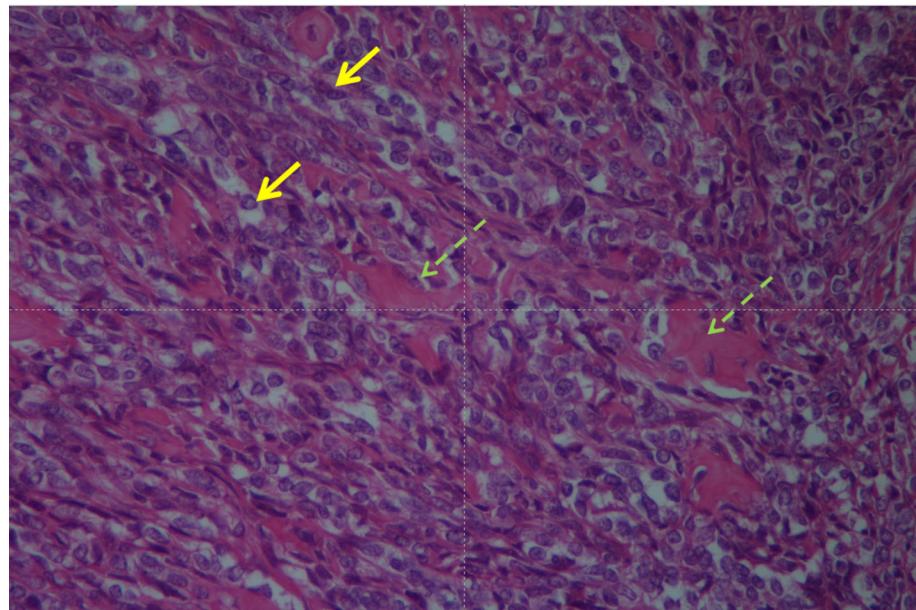
The position of the patient was changed to supine position, and about 8 cm of longitudinal incision was conducted through anterior approach to expose from C5 body to C7 body (Fig. 6) After performing C5–6 and C6–7 discectomy and resecting the left uncovertebral joint, vertebral artery was exposed to anteriorly, clipped, and resected (Fig. 6). In addition, after conducting resection of spine at the right side of C6 body in an oblique way and wide resection of Longus colli, the tumor was removed (Fig. 7a, b). Even after the tumor was removed, wide resection was performed in eight sites including longus colli muscle, C6 lateral margin, anteroinferior margin, posterior margin, posterosuperior margin, posteroinferior margin and inferior margin, and it was found to be margin negative in frozen biopsy. Then, anterior fusion was conducted using demineralized bone matrix and mesh, and skin suture was conducted (Figs. 8, 9).

## Outcome

In the neurologic examination measured after the surgery, symptoms of numbness and poor sense from left thumb to medial forearm were complaint. In the muscular strength evaluation after the surgery, left elbow extension was



**Fig. 10** A  $\times 400$  magnified pathologic photo shows spindle of sporadic mitotic figure or small round type cells (yellow arrow). Relatively abundant and large cytoplasm of cells can be observed (dashed arrow)



measured 4–5, left elbow flexion, wrist flexion, and extension were measured 4+5, and the remaining muscular strength was found to be normal. At day 3 after the surgery, the drainage tube was removed and a neck brace was equipped to walk. At day 14 after the surgery, suture was removed, and the patient was discharged from hospital (Fig. 10). In the followed-up radiograph, the tumor was completely removed, and the instability of joint was not seen. Because of no anticancer drugs suitable for the tumor, adjuvant radiation therapy was conducted in the observation of progression. As a result of observing follow-up CT at a year after the surgery, recurrence findings have not been shown up to now, and the progression of neurologic symptoms has not been shown either.

In the histopathologic examination of removed tumor, spindle of sporadic mitotic figure or cells of small round type were observed, and the patterns of relatively abundant and large cytoplasm of the cells were observed. In the immunohistochemical examination, it was found to be positive to MIC-2 (CD99) (Fig. 10).

#### Compliance with ethical standards

**Conflict of interest** None.

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