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Case Report

Parasagittal resection of multilevel cervical chordoma with autograft duraplasty and unilateral vertebral artery ligation: A case report and literature review☆,☆☆,★

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ABSTRACT

Chordomas are rare tumors and the recommended course of treatment typically entails surgical resection, which presents significant challenges owing to the anatomical location commonly involved and the inherent resistance of these lesions to radiation and chemotherapy. This case report details the experience of a 61-year-old male who underwent a parasagittal resection spanning from C1 to C4. A durotomy was executed to enable en bloc excision of the tumor. Subsequently, a duraplasty procedure was implemented, utilizing autologous muscle fascia grafting. A comprehensive analysis of the pertinent literature was conducted to underscore the key clinical aspects and outcomes related to this topic.

Introduction

Chordomas are malignant tumors originating from embryonic remnants of the notochord, as identified by pathologists [1]. These tumors are most commonly found in the axial skeleton [2].

Chordomas located at the base of the skull account for less than 0.2% of intracranial tumors [3]. The overall incidence of chordomas is 8.4 cases per million patients. They can be

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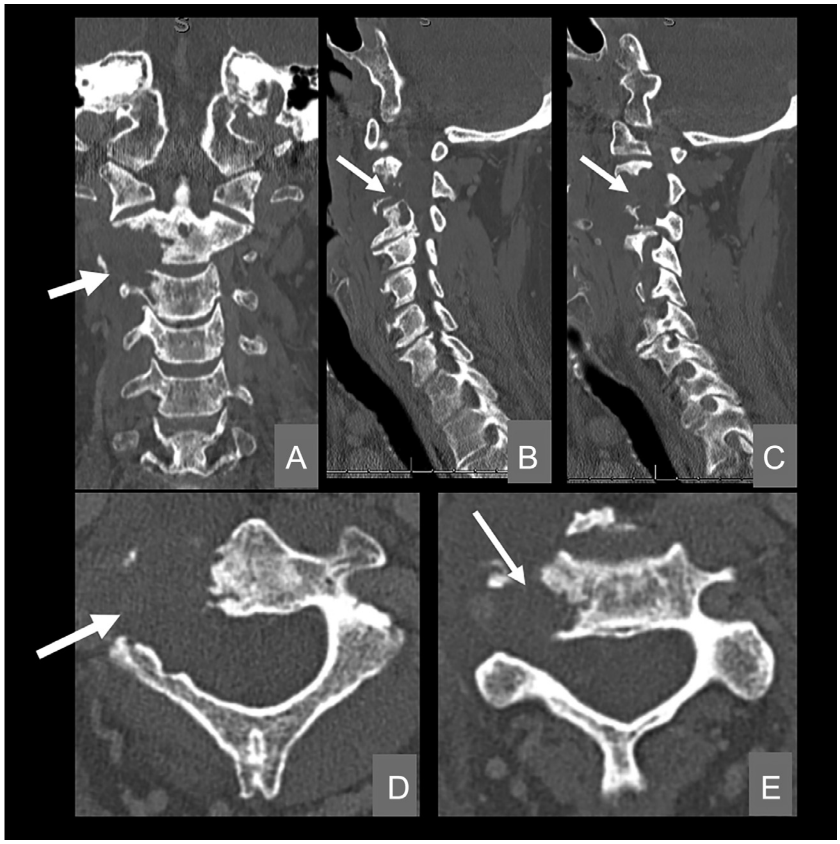

Fig. 1 – Preoperative coronal (A), sagittal (B and C), and axial (D and E) CT scan images showing the lytic bone lesion (white arrows) involving the bodies of C2 and C3, as well as the right C2 and C3 transverse foramina, which are widened.

classified into 3 histological categories: classic (conventional), chondroid, and dedifferentiated [1,3]. The clinical presentation is nonspecific, with vague symptoms, and the most frequent symptom is headache [4].

Computed tomography and magnetic resonance imaging are necessary to determine the tumor's location, bone destruction, and extent of involvement. The recommended treatment typically involves surgical resection, which is often challenging due to the anatomical location usually affected, as well as the fact that these lesions are generally resistant to radiation and chemotherapy [1,5].

En bloc resections of cervical chordomas affecting multiple levels are particularly challenging due to the unique anatomical structure, close relationship with the vertebral arteries, and the spinal cord [5,6].

Most case reports, with or without literature reviews, indicate en bloc resection of cervical chordomas in either a single or 2-stage surgical procedure [7–9]. However, to date, there are few studies evidencing en bloc resection of cervical chordoma involving multiple levels with unilateral ligation of the vertebral artery and none of them described the dural sac resection with autograft duraplasty.

Therefore, the current study aims to report an en bloc resection of cervical chordoma involving multiple levels with unilateral involvement of the vertebral artery, and to conduct a literature review on this topic.

Case presentation

A 61-year-old male presented with neck pain for 3 years without trauma and neurologically intact, without signs of neuropathy or radiculopathy.

The cervical computed tomography (CT) and magnetic resonance imaging (MRI) studies revealed expansive solid and osteolytic soft tissue lesion centered in the right paravertebral region, with lobulated contours determining enlargement of the right neural foramen C2-C3 and erosion of the vertebral body of C2 and C3 in addition to erosion of the transverse process of C2. It shows the extension of the lesion to the interior of the spinal canal determining reduction of its caliber, displacing and compressing the spinal cord. (Figs. 1 and 2).

Staging and preoperative planning

A comprehensive metastatic work-up was negative and a CT-guided needle biopsy of the lesion revealed the diagnosis of chordoma, confirmed on the basis of immunohistochemical studies.

Preoperative studies included an angiography to demonstrate the perviety of both vertebral arteries in case one of them had to be sacrificed. The right vertebral artery was coiled/occluded preoperatively between V2 and V3 vertebral

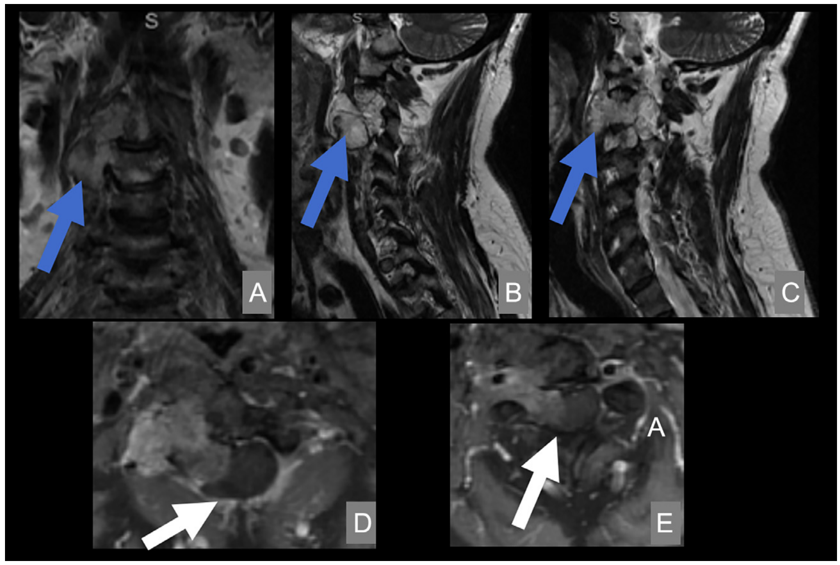

Fig. 2 – Preoperative coronal (A) and sagittal (B and C) MRI images, T2-weighted, revealing an expansive solid soft tissue lesion (blue arrows) situated at the right C2–C3 paravertebral region, associated with bone erosions. Additionally, it displaces and compresses the right vertebral artery. In the axial T1-weighted image with gadolinium (D and E), the extension of the lesion into the interior of the spinal canal is evident, leading to a reduction in its diameter (white arrows).

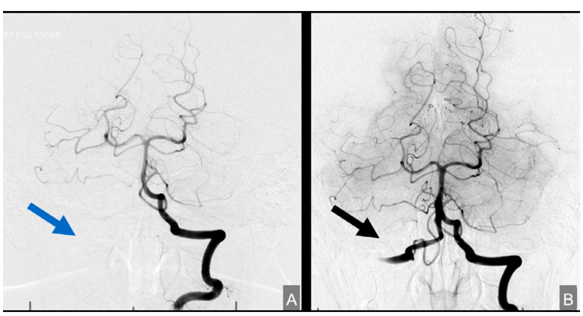

Fig. 3 – Preoperative cerebral angiography, coupled with a balloon test, was conducted. It determined that during the cervical tumor blush, which encompassed the right vertebral artery, there was a negative result in the right vertebral artery occlusion test with regard to neurological deficits. (A) blue arrow - balloon occluded, (B) black arrow - balloon open.

segments, that concluded, during the cervical tumor blush involving the right vertebral artery, negative right vertebral artery occlusion test for neurologic deficits. (Fig. 3).

Preoperative CT and MRI contributed to delineating the location and extent of the tumor and on the basis of imaging findings, Enneking [10] and Weinstein-Boriani-Biagini [11] (WBB) classification system, was performed (Enneking IB; WBB 2-6, A-D, F).

Surgical technique

After general anesthesia was induced, the patient was placed prone. The purpose of the posterior approach (stage 1) is to disconnect the vertebral bodies posteriorly to allow for an en bloc resection anteriorly in the second phase of the procedure.

A midline incision was made to expose the occiput level to the cervical C7 level. Wide laminectomy and facetectomies of posterior elements from C2 to C4. On the right side, the tumor capsule was identified and the plane between the normal soft tissue and the capsule laterally was dissected using Metz scissors, with the dissection continued anteriorly as far as was possible from the posterior approach. The right C2, C3, and C4 nerve roots were tied and cut. Because the right C2, C3, and C4 nerves were already cut, it was easy to access the right vertebral artery.

Subsequently, a posterolateral dissection of the lesion ensued, accompanied by the removal of the dural sac in direct proximity to the tumor. Given the tumor's encroachment on the vertebral artery, a crucial intervention involved the isolation and subsequent clipping of the right VA, carried out between the C1 and C5 vertebrae. Duraplasty was performed with spinal dural repair using autologous muscle fascia grafting (neo dural sac) (Fig. 4). A posterior instrumented fusion was performed from occiput to C7 (Fig. 5).

The second stage performed on the same day involved an extended anterior cervical approach with right paramedian mandibulotomy. A right-sided parasagittal osteotomy through the vertebral bodies of C1 to partial C4 was performed using a high speed bur all the way to the dura, and the tumor involving C1 to C4 was removed en bloc. The entirety of the surgical procedure was conducted within a single operative session.

The multidisciplinary team participating in the surgical procedure comprised spine surgeons, spine surgery fellows, a head and neck surgeon, a head and neck fellow, an anesthesiologist, nurses, a pathologist, a biomedical specialist, an image technician, and an intraoperative neurophysiologist.

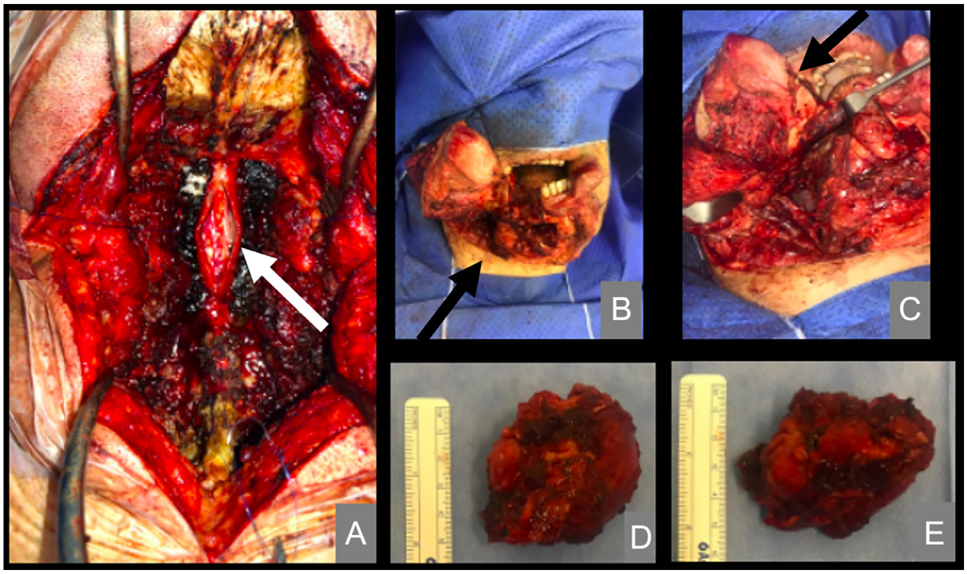

Fig. 4 – Intraoperative images depicting the first (A) and the second stage (B and C) of the surgical procedure are presented. (A) Exposure from occiput to the C7 vertebra, with a wide laminectomy and facetectomies performed on the posterior elements spanning from C2 to C4. Additionally, a durotomy (white arrow) was carried out. (B and C) Anterior cervical approach, complemented by a paramedian mandibulotomy (black arrows). (D and E) surgical specimen.

Fig. 5 – The postoperative CT scan images (A and B) illustrate the parasagittal resection (white arrow) on the right side, as well as the instrumentation (blue arrow) in the posterior occiput-cervical region. Additionally, X-ray images captured in the lateral and anteroposterior views (C and D) display the surgical specimen.

Postoperative course and complications

Intraoperative complication. In the second stage (paramedian approach with mandibulotomy), the antero-lateral cervical anatomical structures were well identified. Despite that, injury to the hypoglossal nerve occurred. It was promptly repaired with a microscope.

Early postoperative complication. The patient was left intubated until postoperative day 2 to allow for surgical swelling to progressively resolve. The first neurological exam after extubation was completely intact. On the third postoperative day, there was a progressive loss of motor strength in the lower and upper right limbs. Through cervical and skull MRI, epidural hematoma in C2 topography on the right and parenchymal hematoma cerebellar with hemorrhagic transformation were diagnosed (Fig. 6).

The cerebellar hemorrhage was treated conservatively and the epidural hematoma was drained with reoperation through a posterior approach. The patient was able to recover his strength and functional status on the right side.

On the following days, during the same hospital admission, the anterior wound developed dehiscence secondary to a salivary fistula. Another 2 revisions of anterior approach to debridement were necessary, followed by diary compressive curative and use of oral drying medication to solve the de salivary fistula.

Late postoperative complication. Probably, due to injury to hypoglossal nerve, the patient developed dysphagia confirmed by swallowing videoendoscopy and classified as severe dysphagia - Grade I according to Dysphagia Severity Scale [12] and with 8 points - contrast turns to glottis with residue in the subglottis but patient does not respond - according to Penetration and Aspiration Scale [13]. Therefore, a gastrostomy was maintained.

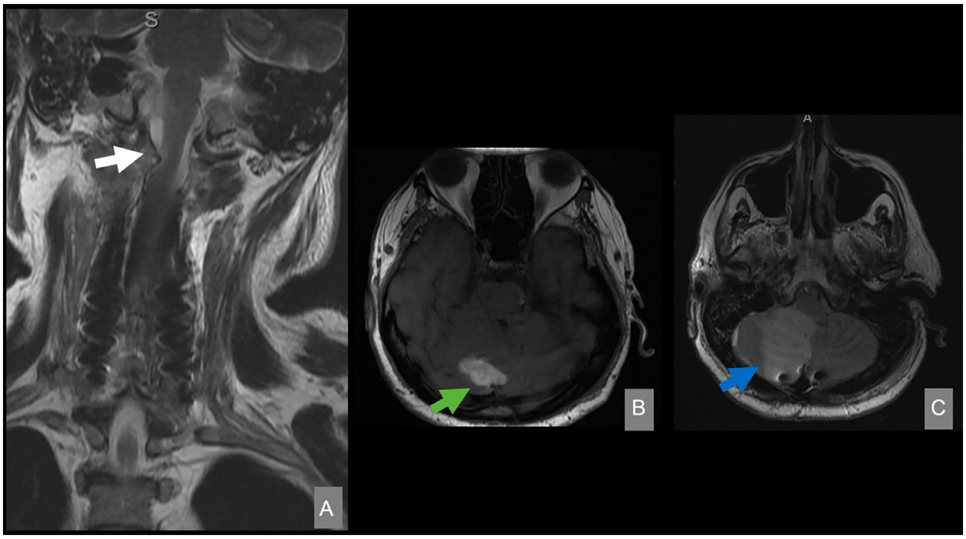

Fig. 6 – (A) MRI coronal view showing an epidural hematoma at the C2 level (white arrow). (B and C) MRI axial view demonstrating a cortico-subcortical swelling zone (blue arrow) that impacts the posterior and inferior aspects of the right cerebellar hemisphere. A hematoma (green arrow) measuring 3.3 x 1.7 cm is observed, contributing to the compression of the fourth ventricle.

Fig. 7 – (A–F) Clinical images showing the surgical scars (white arrows) after 51 months and the alignment of the spine (green arrows). (G–I) Cervical X-rays demonstrating the instrumentation (blue arrows) that appears to be intact.

The patient was hospitalized on a single occasion for the specific surgical procedure and encountered complications throughout the intraoperative, early postoperative, and late postoperative phases as previously described.

MRI obtained at 51-months postoperatively did not show any residual or recurrent disease. The cervical X-ray reveals the apparent integrity of the instrumentation, and the surgical scars exhibit thorough healing (Fig. 7).

Literature review

Study eligibility criteria: the review searched all types of studies that addressed en bloc resection of cervical chordomas and vertebral artery ligation. We excluded studies unrelated to this topic, not retrievable in full texts, or not in the English language.

Information source: the search strategy was based on the Population Intervention Comparison Outcome (PICO) method [14] {P: multilevel cervical chordoma patients; I: en bloc resection with unilateral vertebral artery ligation; C: none; O: postoperative surgical complications} and included all articles published in English without date restrictions. Articles were searched in the PubMed database with the keywords "chordoma, vertebral artery," "chordoma, en bloc resection," "cervical chordoma, vertebral artery," cervical chordoma, en bloc resection. The last date when the search was done was on September 19, 2023.

Search strategy: two researchers (MC, LS) conducted the search separately and the final selection was made by cross-checking the abstracts identified by both. A 3-step search strategy was used. An initial limited search was undertaken, followed by an analysis of the keywords contained in the title and abstracts across all included databases. A second search involved checking the abstracts against the inclusion criteria. Finally, the search was complemented with searches of references and a list of included studies.

Data extraction and synthesis: the retrieved studies were initially screened by reading the title and abstract. Two reviewers (MC, LS) independently extracted data from each study, including general information, data of publication, study design, participants' characteristics, outcome measures, and key results. A table approach was used to synthesize the study findings, outcomes, and clinical features.

Results

The search identified 377 records. All of them were examined and 15 articles were eligible for inclusion. Table 1 synthesizes the study findings, outcomes, and clinical features.

Discussion

The literature supports the notion that en bloc resection represents the sole treatment modality associated with extended disease-free survival for patients afflicted with chordomas. The prognosis of chordoma is profoundly influenced by the appropriateness of the initial treatment, as subsequent interventions following recurrence invariably yield unsatisfactory outcomes in the medium term [27]. It is important to note that the treatment of chordoma is essentially a single, crucial surgical intervention, with the initial attempt being indisputably the most effective [28].

Hsieh et al. [8] in their research findings underscored the significant impact of the specific type of en bloc resection on patient outcomes, as demonstrated in their analysis of 5 cases. Indeed, their investigation elucidated that "en bloc" resection represents an oncological principle predicated on the complete removal of the tumor as a single intact unit, without any breach of its pseudocapsule.

As an alternative approach to multilevel spondylectomy for the management of multilevel cervical chordomas, a staged parasagittal osteotomy technique was used to achieve en bloc removal of multiple segments of the cervical spine affected by chordoma. It is noteworthy that while these procedures do not amount to total spondylectomy, they remain in alignment with the fundamental oncological principle of achieving en bloc excision [19].

In practice, achieving en bloc resection with wide margins in the cervical spine is a formidable task, necessitating the inclusion of not only the dura, nerve roots, and vertebral arteries but also extensive soft tissue in the margin. To meet these requirements, after complete lesion excision and intraoperative confirmation that the tumor adhered to the dural sac without intrathecal invasion, was opted to encompass the dura, nerve roots, and the right vertebral artery in the resection.

Surgery continues to be the primary treatment modality for chordomas, with a paramount emphasis on attaining wide margins. Surgical techniques focusing on en bloc resection with tumor-free margins have demonstrated favorable outcomes in terms of local control and long-term prognosis for chordomas arising in the thoracic and lumbar spine. Nevertheless, the cervical spine poses unique challenges in achieving a genuine en bloc resection [9].

The primary challenge in achieving a comprehensive en bloc resection of cervical chordomas lies in the proximity of vital structures such as the vertebral artery and nerve roots. It is important to recognize that sacrificing the vertebral artery carries the risk of causing a stroke, particularly when the tumor is closely associated with the dominant vertebral artery and spinal cord ischemia due to the variability of the radiculomedullary branches [29].

In this report, a thorough preoperative assessment to determine the feasibility of sacrificing the vertebral artery assumes critical importance before contemplating an en bloc resection. The potential neurological morbidity stemming from vertebral artery sacrifice can be profoundly debilitating. Therefore, conducting a balloon occlusion test to assess collateral circulation should precede any decision to sacrifice the artery. Fujita et al. [15], pioneered the use of a preoperative vertebral artery occlusion test before undertaking en bloc resection of a cervical chordoma. During their surgical procedure, they ligated the artery prior to the removal of the C5 vertebra. Similarly, Rhines et al. [17] employed temporary aneurysm clips on the patient's right vertebral artery intraoperatively, while closely monitoring somatosensory evoked potentials for a duration of 30 minutes before proceeding with the intraoperative ligation and transection of the artery, both above and below the tumor site.

Cloyd et al. incorporated their neurointerventional service to preoperatively occlude the right vertebral artery in a specific case. In their review, Hsieh et al. [7] recommended the use of cerebral angiography and a temporary balloon occlusion test, although they did not elaborate on preoperative embolization.

In a case series by Wang et al. [30], the sacrifice of the vertebral artery was performed in 3 of 14 patients without reported complications; however, there was no mention of preoperative testing.

Despite obtaining negative results for neurological deficits in the balloon occlusion test of the right vertebral artery, a subsequent development of cerebellar ischemia within the territory of the posterior-inferior cerebellar artery was observed. Govsa et al. [31] meticulously noted the variability in the vertebral artery's termination, whether as a posterior inferior cerebellar artery or as the anterior spinal artery with a unilateral origin (type II). This observation may provide insight into the occurrence of the ischemic insult.

Rhines et al. [17] reported several complications, including persistent swallowing difficulties lasting for 1 year postoperatively, migration of the caudal graft and plate, erosion of the posterior pharyngeal wall with exposed cervical implants leading to the need for a ra-

Table 1 – Studies with multilevel cervical chordoma and vertebral artery ligation.

Author/year	Type of study	Number of patients	Age/ Sex	Level involved	Surgery - Staging	Resection type	Follow-up period	Type of vertebral artery occlusion/ ligation	Local Recurrence	Complications
Fujita et al. (1999) [15]	Case report	1	16, male	C5–C6	1	En bloc	30 mo	Open ligation	No	Hypesthesia; motor weakness
Carpenter et al. (2001) [16]	Case series	36 (4 with vertebral artery ligation)	11-36; 16 female, 20 male	Most upper cervical; 8 C2-C6; 1 C1-T1	1 - 2	En bloc	1-11 y	Open ligation	Yes (40%)	Transient Horner syndrome; nerve weakness; cardiopulmonary arrest during transoral approach
Rhines et al. (2005) [17]	Case report	1	54, male	C2–C4	2	En bloc - Marginal	12 mo	Open ligation	No	Several weeks ventilatory support; swallowing difficulties; graft and plate migration; pharyngeal erosion and implants exposition
Bailey et al. (2006) [18]	Case report	1	50, male	C1–C3	3	En bloc - Marginal	24 mo	Open ligation	No	Septic shock; acute respiratory distress syndrome; swallowing difficulties
Chou et al. (2009) [19]	Case series	3	59, male	C3–C4	2	En bloc – marginal	30 mo	Open ligation	No	Aspiration pneumonia; dysphagia; wound dehiscence
			62, female	C3–C5	2	En bloc - marginal	16 mo	Open ligation	No	Seizure
			61, male	C2–C4	2	En bloc - wide	12 mo	Open ligation	No	Pneumonia; dysphagia
Cloyd et al. (2009) [7]	Case series	1	60, female	C3–C5	2	En bloc - intralesional	12 mo	Endovascular	Yes	Pneumonia; Respiratory difficult; thrombosis of the tracheotomy; Upper extremity motor weakness
Jiang et al. (2009) [20]	Case report	1	26, female	C2–C3	1	En bloc - Wide	36 mo	Open ligation	No	Meningocele
Hsieh et al. (2011) [8]	Case series	2	52, male 32, male	C2–C4 C2–C3	2 2	En bloc – wide En bloc - wide	71 mo 22 mo	Open ligation Open ligation	No	Upper extremity motor weakness; postsurgical infection; pseudoarthrosis / instrumentation failure
Vassal et al. (2012) [21]	Case report	1	45, female	C2–C4	3	En bloc - wide	24 mo	Open ligation	No	Respiratory difficult
Guppy et al. (2013) [9]	Case report	1	49, female	C2–C3	3	En bloc -Intralesional	20 mo	Endovascular	Yes	Motor weakness; pharynx dehiscence
Weil et al. (2015) [22]	Case report	1	46, female	C2–C3	2	En bloc - Wide	40 mo	Open ligation	No	Respiratory difficult; Upper extremity weakness; dysphagia
Xiao et al. (2016) [23]	Case series	5 (1 with vertebral artery ligation)	67, male	C4–C5	1	En bloc -Marginal	21 mo	Open ligation	No	Respiratory difficult; Upper extremity weakness
Mesa-Quesada et al. (2017) [24]	Case report	1	33, male	C2–C4	1	En bloc -Intralesional	2 d	Endovascular	Yes	-
Molina-Martínez et al. (2021) [25]	Case report	1	40, male	C3–C4	3	En bloc - Wide	2 wk	Open ligation	No	-
Pinter et al. (2022) [26]	Case report	1	67, male	C1–C3	2	En bloc - Wide	132 mo	Endovascular	No	Dural tear; cerebellar hemorrhage; local abscess; cerebrospinal fluid leak;

dial artery forearm free flap, as well as the need for a gastrostomy.

In this report, numerous complications transpired, encompassing those associated with the anatomical location of the tumor (cerebellar ischemia likely attributable to vascular abnormalities in the cervical spine), complications arising from the surgical approach (salivary fistula), and those arising from inherent surgical risks (injury to the hypoglossal nerve). In addition to the aforementioned complications, it is noteworthy that during the 51-month follow-up postoperatively, the patient demonstrated enhanced well-being. The individual was capable of ambulating without reliance on any assistive devices and engaged in routine daily activities normally. Nevertheless, a lasting consequence persisted, the severe dysphagia, for which the use of a gastrostomy remained necessary.

During the tumor board discussions held both prior to and a few weeks subsequent to the surgery, wherein the oncologist, pathologist, spine surgeon, radiologist, and radiation therapy specialist were in attendance, the initially devised treatment strategy did not incorporate either radiation therapy or chemotherapy. This is partially attributed to the achievement of negative margins during the surgical procedure. Following the 51-month follow-up, the established course of treatment involves periodic annual visits, accompanied by relevant examinations tailored to oncologic surveillance needs.

Conclusion

The management of chordoma fundamentally revolves around a singular, pivotal surgical procedure, wherein the initial endeavor undoubtedly proves to be the most efficacious. In this particular case, we underscore the significance of promptly diagnosing the tumor and emphasize the imperative of seizing the opportunity, whenever feasible, to perform an en bloc resection in order to enhance disease-free survival.

Patient consent

Patient consent has been obtained.

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