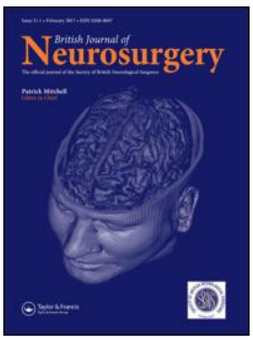

British Journal of Neurosurgery

ISSN: (Print) (Online) Journal homepage: https://www.tandfonline.com/loi/ibjn20

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To cite this article: Reza Mousavi, Majid Reza Farrokhi, Keyvan Eghbal, Javad Safaee & Amir Reza Dehghanian (2021): Reconstruction of C1 lateral mass with an expandable cage in addition to vertebral artery preservation: presenting two cases, British Journal of Neurosurgery, DOI: 10.1080/02688697.2021.1978393

To link to this article: https://doi.org/10.1080/02688697.2021.1978393

Published online: 17 Sep 2021.

BRITISH JOURNAL OF NEUROSURGERY
https://doi.org/10.1080/02688697.2021.1978393

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REVIEW ARTICLE

Reconstruction of C1 lateral mass with an expandable cage in addition to vertebral artery preservation: presenting two cases

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ABSTRACT

Purpose: C1 lateral mass reconstruction is recommended, in cases of instability caused by tumor involvement or extensive C1 lateral mass resection. However, because of the anatomical complexity of the area and, most importantly, the proximity to vertebral arteries, few cases of reconstruction have been reported to date. The purpose of this report is to present technical details of C1 lateral mass reconstruction in conjunction with vertebral artery preservation from a posterior approach.

Methods: Two cases of one stage craniovertebral junction instrumentation and C1 lateral mass reconstruction in conjunction with vertebral artery preservation from a posterior approach are presented.

Results: In both cases of extensive resection of lateral mass due to tumor involvement, an expandable cage was used for C1 lateral mass reconstruction, which has been used only in one patient in literature. Complementary pathological examinations of the two cases indicated two rare tumors that had been reported in the upper cervical region so far. The first case became an unknown origin metastatic cancer and the second was reported to be a primary non- Hodgkin lymphoma.

Conclusions: C1 lateral mass reconstruction with an expandable cage together with VA preservation is recommended in cases of extensive C1 lateral mass resection to increase the total strength and to shorten the length of the posterior device and probably better fusion. The expandable cage is preferred because of safer placement under compression instead of the lateral mass.

ARTICLE HISTORY
Received 28 December 2020
Revised 5 July 2021
Accepted 6 September 2021

KEYWORDS
CVJ instability; C1 lateral mass reconstruction; expandable cage; vertebral artery preservation; unknown origin metastasis; non-Hodgkin lymphoma

Introduction

The first cervical vertebra is unique because of lacking the vertebral body.[1,2] In the case of tumoral involvement or extensive surgical resection of this important bony structure, its reconstruction is recommended.[3–9] However, because of the anatomical complexity of the area and, most importantly, the proximity to vertebral arteries, few cases of reconstruction have been reported to date. Two cases that we are reporting here are the second and third in the related literature, for whom an 'expandable cage' has been installed for C1 lateral mass reconstruction.[8] In these operations, we also preserved the ipsilateral vertebral artery (VA).

Case reports

Case 1

A 33-year-old woman presented with progressive mechanical neck pain since 1 month earlier. On physical examination, the patient exhibited local tenderness in the upper cervical region. No neurological deficit was detected. Cervical magnetic resonance imaging (MRI) revealed a heterogeneous enhanced lesion of right C1 lateral mass (C1 LM) with some extension to the anterior arch and minimal invasion to the spinal canal without cord compression. A cervical 3 dimensional computed tomography (CT) showed an osteolytic $20 \times 23 \times 30$ mm right C1 LM lesion (Figure 1). CT angiography (CTA) revealed two anatomic variations; first, the left VA was originated from the aortic arch and was hypoplastic, andsecond, the right VA was dominant. The right VA size was significantly larger than the left. Abnormal vascularity of the tumor was not apparent.

Case 2

A 52-year-old man presented with progressive severe mechanical neck pain and tenderness, and limitation of neck motion since 2 months earlier. Cervical MRI and CT scan revealed a huge retropharyngeal mass ($6 \times 3.5 \times 2$ cm) with extension to and destruction of right C1 LM. They also demonstrated homogenous enhancement and extradural extension to the spinal canal (Figure 2). CTA revealed an equal size of both VAs while the right VA was encased by the tumor but it was patent .No critical anatomical variation was observed on CTA.

Preoperative tests for both cases such as lab tests, chest and abdominal CT, and positron emission tomography (PET) scan did not reveal any metastatic origin. Also, results of a closed CT-guided biopsy were unremarkable.

Our operative planning for both cases was tailored as one stage posterior occipitocervical fixation in addition to intralesional resection of lesions, followed by right C1 LM reconstruction using an expandable cage and VA preservation.

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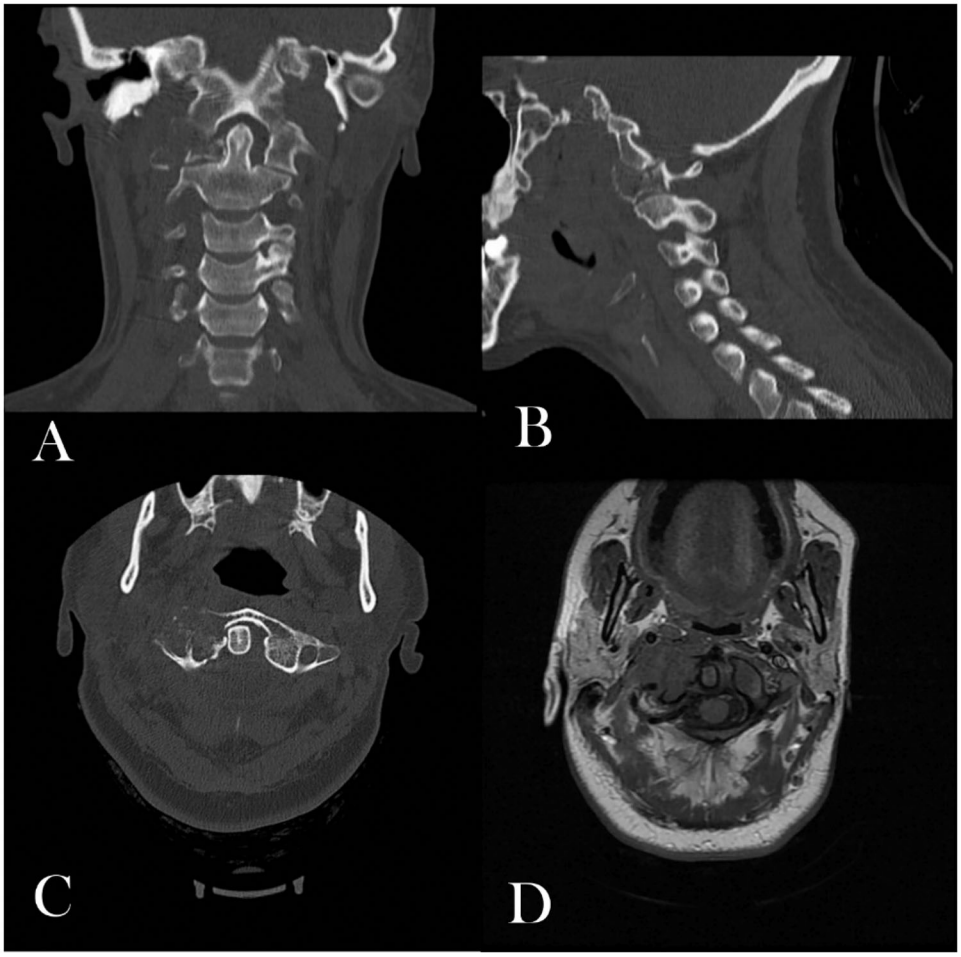

Figure 1. Case 1. Preoperative tomogram (A, B, C) and MRI (D) showing an osteolytic lesion of right C1 lateral mass.

Technical details

Our operation technique for both cases was similar and included four main steps from a posterior approach. In the first step (occipitocervical instrumentation), we placed a keel occipital plate, a left-sided C1 LM, and bilateral C2 pedicle screws. In the second step (dissecting and releasing VA), we sacrificed the right C2 root proximal to its ganglion and venous plexus around it. The right half of the C1 posterior arch and its pedicle was removed. We identified and dissected the third part of the right VA on the dorsal groove of the posterior C1 arch. Then we encountered the KEY point of the procedure- i.e., releasing VA from its most critical tethered point by unroofing C1 transverse foramen. Then, VA was skeletonized from C2 transverse foramen to C1 transverse foramen. In the third step (LM resection), using Kerrison rongeurs and curettes, we removed the erosive lesion of C1 LM by central decompression and piecemeal fashion. In both cases, the tumor had a soft consistency and did not have considerable bleeding. Extradural portion of the tumor was also removed. In the fourth step (reconstruction), after decortication of occipital condyle (OCO) and C2 superior articular process, we performed reconstruction of the C1 LM under fluoroscopy using an expandable titanium cage (12–17 mm) filled with autograft. In order to have the best control of the cage during the insertion and expansion, we followed three strategies; first, the cage was applied fully unexpanded while entering the corridor; second, we placed it upside down, so its basic cylinder perched on the OCO and expandable cylinder settled on C2 SAP (in order to increase the workspace for expander handle), and; third, the cage was expanded to the maximum length that was allowed under compression.

Postoperative period

Postoperatively, both patients had a non-complicated recovery course. Apparently, their pain was resolved, and no neurological deficit was generated. On the second day, a CTA was requested for each case to ensure that the VA pathway was left open and was not compromised by the cage (Figure 3). In the 3rd month of follow-up, both patients were pain-free with favorable control radiographs (Figure 4).

Discussion

Two-fifth of the Atlas ring has been made by the LMs.[1] Its average midportion (central) length is 16.82 mm (SD 1.0), width 16.06 mm (SD 0.91), and height 15.68 mm (SD 0.98).[2] The transverse ligament tubercles on the medial aspect of the LMs serve as the lateral insertion point of the ligament.[2]

Only $25-40.8\%$ of normal population have equal-diameter left and right VAs.[1,2] Left VA in $35.8-50\%$[1,2] and right VA in $23.4-25\%$[1,2] of cases are dominant. The left VA is hypoplastic in 5.7% and absent in 1.8%.[2] The right VA is hypoplastic in 8.8% and absent in 3.1%.[2] In case 1, we dealt with a right dominant VA. Left VA was hypoplastic, so preoperative embolization of

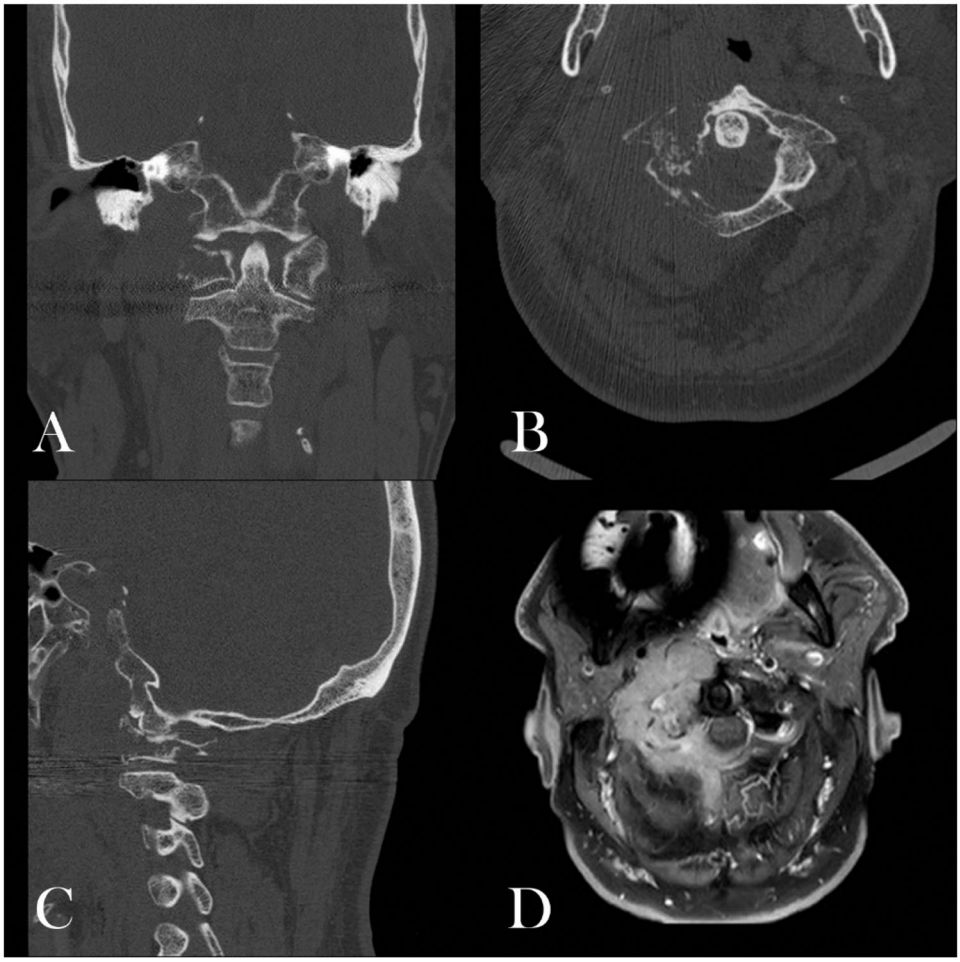

Figure 2. Case 2. Preoperative tomogram (A, B, C) and MRI (D) showing a large destructive lesion of right C1 lateral mass with extension to retropharyngeal space.

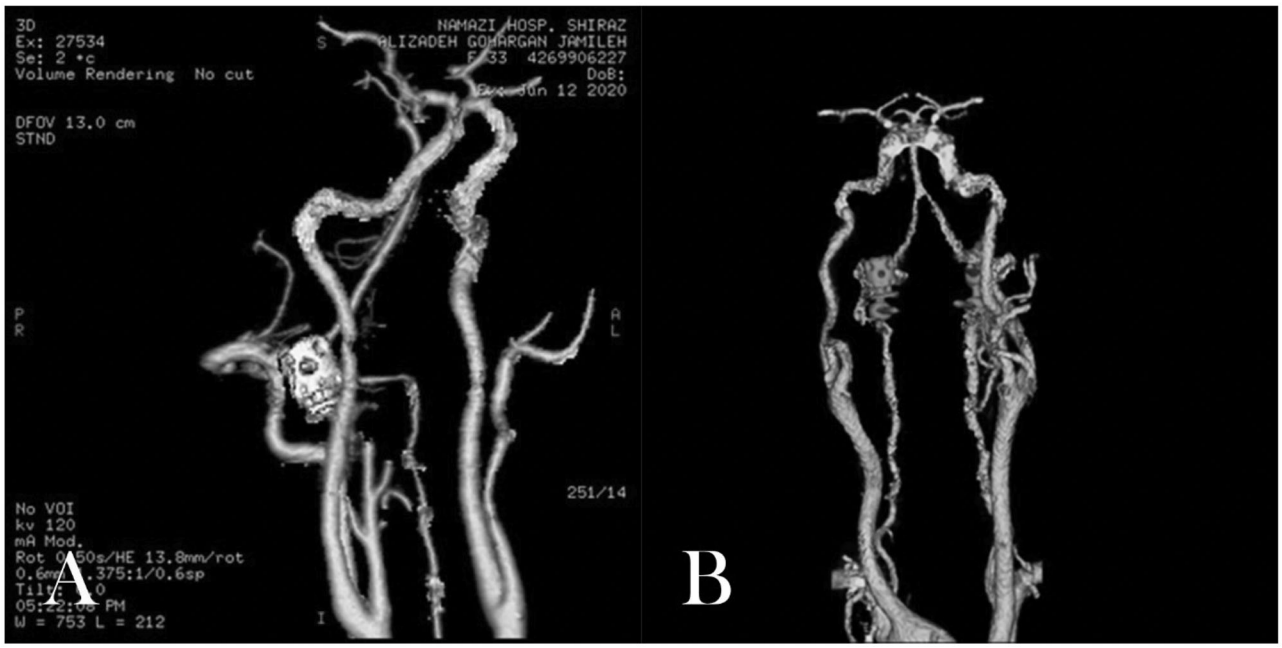

Figure 3. Postoperative computed tomographic angiogram of case 1 (A) and case 2 (B) revealed VAs have been left patent.

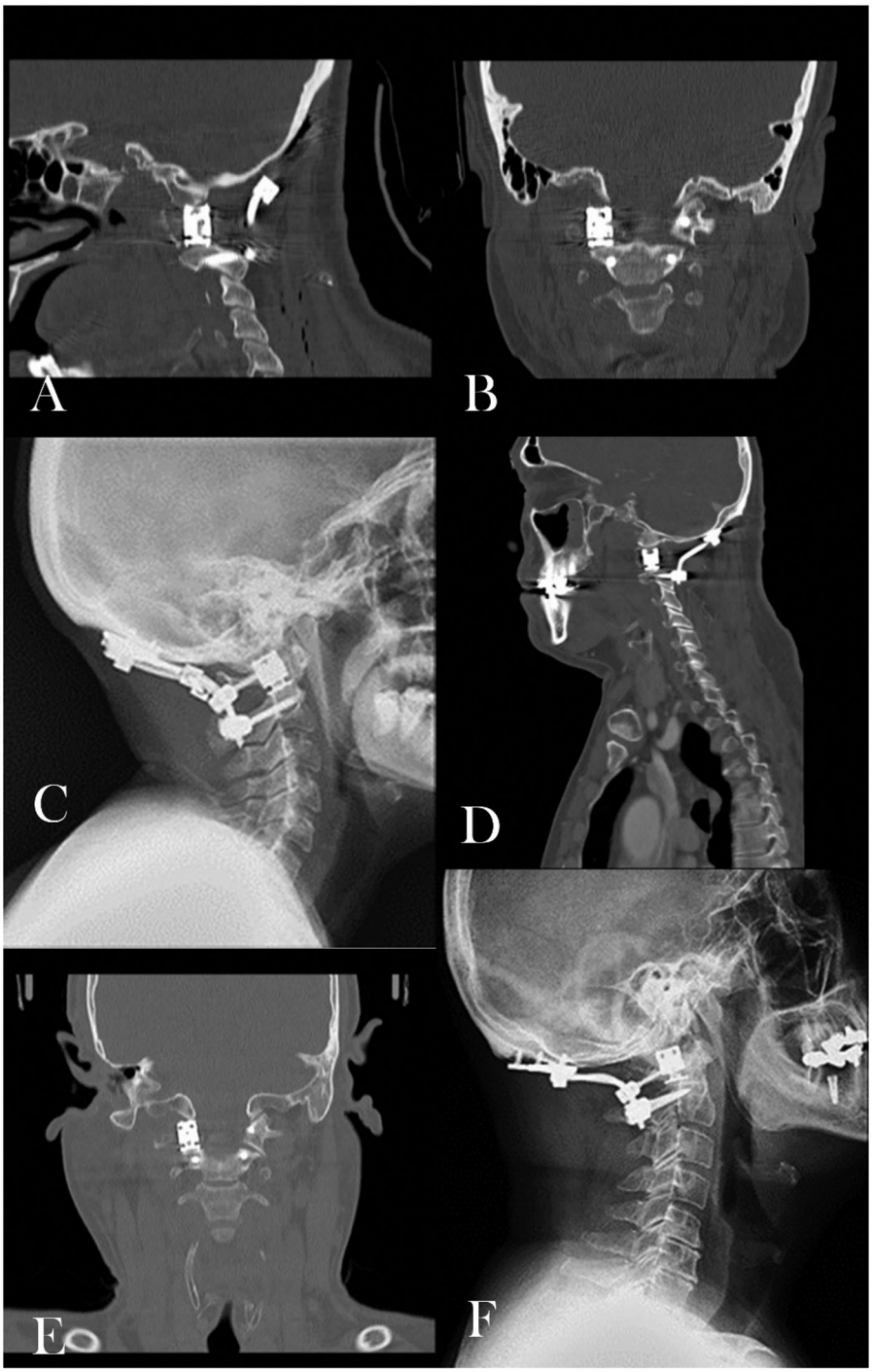

Figure 4. Postoperative 3 month follow up: CT scans (A, B) and lateral X-ray (C) of case 1. CT scans (D,E) and lateral X-ray (F) of case 2.

the right VA was not possible, and its preservation was critical. In our 2nd case, both VAs had equal diameter. The incidence of VA anomalies is about 3.5–19.2%.[1,2] It is important to be aware of following anatomical variations in the operation corridor of the C1 LM reconstruction or C1 LM screw insertion:

A. Ponticulus posticous (up to 15.6%) – i.e. bony covering of the V3 segment of VA on the C1 posterior arch.[2]
B. Persistent primitive first cervical intersegmental artery (unilaterally in 3.8% and bilaterally in 0.8%)[2]
C. An extradural origin of the posterior inferior cerebellar artery (PICA) (5–20%) which can arise from V3 or even V2.[2]
D. The posterior spinal artery (PSA) which may originate from V3 in 46% or from an extradural PICA.[2]

According to the spinal instability neoplastic score (SINS),[10] any osteolytic lesion of C1 LM presenting with pain will have a minimum score of 7 (3 for junctional location, 2 for pain, and 2 for lytic lesion), which means impending or overt instability requiring surgery. SINS of our both cases was 12. Traditionally, posterior occipitocervical instrumentation with multiple anchoring choices is used to restore the stabilization of CVJ. On the other hand, the load sharing properties of the atlas need to be noticed. Atlas is a unique and unusual vertebra, lacking a body.[2] The weight-bearing vector across the C1 passes along C1 LM of

Table 1. Literature review of C1 lateral mass reconstruction with a titanium cage.

No	Study	publication	Number of cases	Approach	Pathology	VA preservation
1	Wang et al.	2009	1	Posterior	ABC	Occluded
2	Chung et al.	2012	1	Lateral	Osteosarcoma	Occluded
3	Bobinski et al.	2014	2	Posterior	1. Multiple myeloma 2. Angiosarcoma	Preserved
4	Winking	2014	1	Lateral	Plasmocytoma	Preserved
5	Peciu-Florianu et al.	2016	1	Posterior	Osteoblastoma	Preserved
6	Bradley et al.	2016	1	Posterior*	Eosinophilic granuloma	Preserved
7	Neva et al.	2017	1	Posterior	ABC	Preserved
8	Mousavi et al. (present study)	2020	2	Posterior*	1. Unknown origin metastasis 2. Primary non-Hodgkin's lymphoma	Preserved

*Expandable cage is used.

each side, directly from OCO to C2.[1] Known relevant indicators of instability are:

A. Tumoral involvement or surgical resection of bony elements of CVJ can lead to instability: Axial rotation >8 of O-C1 to one side indicates instability.[11]
B. Craniovertebral junction stabilization should be considered if more than 50%[12,13] or 75%[7] of one occipital condyle is resected.
C. C1 LM resection may lead to instability because of transverse ligament (TL) incompetency.[5,7]
D. Resection of C1 LM beyond 'prime meridian' leads to the downward and posterior slope of OCO over C1.[7]
E. Bilateral tumoral involvement or resection of C1 LMs leads to the complete destruction of the weight transfer pathway;[7] therefore, reconstruction is recommended.

Histopathological evaluation of both tumors shows undifferentiated discohesive cells with no clear cancer lineage differentiation, so immunohistochemistry (IHC) is mandatory to make a more precise diagnosis. In our case 1, the immunohistochemical study revealed CK7+/CK20- cells, in favor of metastatic carcinoma. Primary origins of CK7+/CK20- metastatic carcinomas could be lung, breast, female genital tract, thyroid, pancreato-biliary system, and a subset of gastric adenocarcinoma as well as a subset of urothelial carcinoma.[14] about 70% of end-stage cancers show vertebral bone metastasis.[15] Amongst these patients, atlantoaxial involvement is about 0.5% of all spinal metastases.[16] The most common site of primary atlantoaxial metastasis is the lung followed by nasopharynx, liver, and thyroid, according to previous studies.[17] In our 2nd case, LCA immunoreactivity confirms the diagnosis of the hematopoietic origin of malignancy, and further evaluations led to the diagnosis of germinal center-type (GC-Type) diffused large B cell lymphoma (DLBCL). Reportedly, non-Hodgkin's lymphoma of the vertebra is quite rare and accounts for 0.1–3.3% of all the malignant tumors of the vertebra.[18] GC-type DLBCL has a more favorable prognosis compared with non-GC-type, and fortunately, this type is more common among vertebral DLBCLs.[19]

Literature is still scarce about C1 LM reconstruction. In searching databases, we found that most of the related articles are small case series. Reconstruction of C1 LM is also called occipitoaxial spinal interarticular stabilization (OASIS) in some articles.[5,7] In total, only eight cases of C1 LM reconstruction had been reported to date, all of which have been done unilaterally except one: a bilateral recurrent C1 osteoblastoma..[7] Preoperative occlusion of the ipsilateral VA has been performed in two of these cases.[3,4] Intraoperative Navigation (ION)[9] or micro-Doppler[5] can be used to confirm that VA remains open. In most of those cases, titanium cages had been used (Table 1). In addition to our two cases, "expandable cage" has been used only in one other case.[8] Considering the literature and our reported cases, the following points can be highlighted:

1. For unstable pathological condition of C1LM, in addition to posterior occipitocervical fixation, C1LM reconstruction is recommended in the following cases: resection of C1 LM beyond meridian (i.e. more than 50%), and bilateral tumoral involvement or resection of C1 LMs.
2. VA preservation is recommended for two reasons. First, to avoid neurological deficits. The risk of neurological deficit following VA sacrifice is reported as 6% in the literature.[5] Second, to reduce the number of levels involved in fusion. In cases that ipsilateral VA is occluded preoperatively, pedicle screw placement of contralateral C2 is contraindicated because of the risk of injury to only one remaining VA. In these cases, elongation of occipitocervical fixation to C3 and C4 LM mass is needed.[3]
3. C1 LM reconstruction can be done from a posterior, direct lateral, or modified far lateral approach. We believe that the posterior approach is relatively safe and recommended for several reasons. First, spine surgeons are well familiar with it. Second, in the posterior approach, contrary to Winking,[6] VA releasing can be performed early by dissection along its groove on the posterior arch of the C1 and by unroofing C1 transverse foramen. Third, both stabilization and reconstruction are performed from one posterior approach, while the lateral approaches necessarily include two stages: the lateral for reconstruction and the posterior for stabilization.[4,6] Fourth, in lateral or modified far lateral approachs, before accessing C1 transverse foramen, the internal jugular vein and accessory nerve are on the way.[6] As a result, the risk of damage to the main arteries and lower cranial nerves, in addition to VA injury, will be added to these approaches.
4. Spacers like titanium mesh cages for C1 LM reconstruction, by load sharing and reducing pressure on posterior stabilization devices, increase the fusion rate and decrease the likelihood of instrumentation failure and also reduce the length of posterior fusion.[5] The advantages of using an expandable cage include more secure embedding due to initial and final size adjustments.

Conclusion

C1 LM reconstruction with an expandable cage together with VA preservation is doable and is relatively safe if some critical tips are followed. Preoperative CTA or selective angiography is mandatory to find about the VA course and its branches and probable variations. The key point of the procedure is VA untethering in C1 transverse foramen. We recommend C1 LM reconstruction to increase the total strength and to shorten the length of the posterior device and probably better fusion. In our opinion, the merits of using expandable cage for C1 lateral mass reconstruction could be its being easily and safely inserted as

well as its lower chance of failure, which is due to its better fixation and insertion. As to the drawbacks, the low volume of central cylinder of cage for bone graft impaction can be taken into account.

Acknowledgments

The authors would like to thank Dr. Elham Nasiri for thoroughly editing the manuscript and Ms. Hosseini from the Shiraz Neuroscience Research Center for her help. This article has no source of funding.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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