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A Radiologists' Guide to En Bloc Resection of Primary Tumors in the Spine: What Does the Surgeon Want to Know?

E. Smith[1] G. Hegde[1] M. Czyz[2] S. Hughes[2] S. Haleem[2] M. Grainger[2] S. L. James[1] R. Botchu[1]

[1] Department of Musculoskeletal Imaging, Royal Orthopaedic Hospital, Birmingham, United Kingdom
[2] Department of Spinal Surgery, Royal Orthopaedic Hospital, Birmingham, United Kingdom

Address for correspondence Rajesh Botchu, Department of Musculoskeletal Radiology, The Royal Orthopedic Hospital, Bristol Road South, Northfield, Birmingham, B31 2AP, United Kingdom (e-mail: drbrajesh@yahoo.com).

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Abstract

En bloc resection in the spine is performed for both primary and metastatic bone lesions and has been proven to lengthen disease-free survival and decrease the likelihood of local recurrence. It is a complex procedure, which requires a thorough multi-disciplinary approach. This article will discuss the role of the radiologist in characterizing the underlying tumor pathology, staging the tumor and helping to predict possible intraoperative challenges for en bloc resection of primary bone lesions. The postoperative appearances and complications following en bloc resection in the spine will be considered in subsequent articles.

Keywords
- en bloc
- spine
- primary tumors

Introduction

En bloc excision in the spine is a complex procedure with high morbidity and mortality rates.[1] It is not a commonly performed surgery, and a thorough multidisciplinary approach to each case is imperative.[2] Imaging cannot only help characterize the underlying tumor pathology but has a pivotal role in assessing the position and extent of the tumor to accurately stage the tumor and assess suitability for resection.

This article will discuss the indications for en bloc excision in the spine and the oncological staging of spinal tumors. The surgical staging of spinal tumors will also be described along with an explanation of how this affects the surgical approach. The aim is to allow the radiologist to gain a greater understanding of the pre- and intraoperative considerations of the surgeon to provide relevant reports, which will optimize preoperative planning.

En Bloc Resection

The term en bloc resection (EBR) is given to the procedure that attempts to remove the entirety of a tumor in one piece along with a surrounding margin of healthy tissue.[1] The oncological principles involved in en bloc tumor resection were initially well established for bone tumors of the limbs, and subsequently, these principles inspired a standardized approach to tumors of the spine.

Experiences of the complete removal of the vertebra in spinal tumors were first described by Stener in 1989.[3] A further popular standard technique was described by Roy Camille and Tomita.[4] This approach, however, was only really suited to the thoracic spine and was only appropriate if the tumor was positioned centrally and contained entirely within the vertebral body. There was an obvious need for a staging system for spinal tumors that would describe tumor extent and position to allow for a standardized surgical approach

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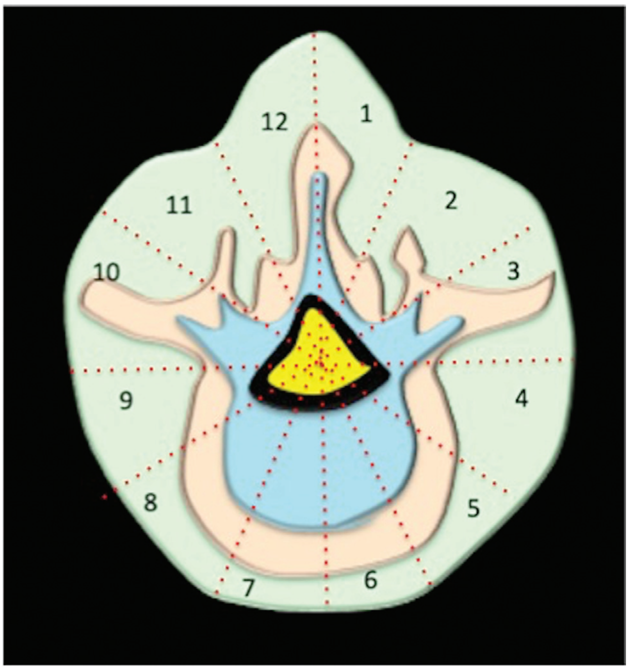

Fig. 1 Schematic of vertebra showing Weinstein-Boriani-Biagini zones 1–12, extraosseous soft tissue (**a**—green), intraosseous superficial (**b**—light orange), deep intraosseous (**c**—blue), extraosseous, extradural (**d**—black), extraosseous intradural (**e**—yellow).

Table 1 WBB surgical staging system

A—Soft tissues (extraosseous)
B—Superficial (intraosseous)
C—Deep (intraosseous)
D—Extradural
E—Intradural

Abbreviation: WBB, Weinstein-Boriani-Biagini.

based on oncological principles. This was the reason for development of the Weinstein-Boriani-Biagini (WBB) staging system.[1] (►**Fig. 1**, ►**Table 1**)

Which Tumors Are Appropriate for En Bloc Resection?

Due to the significant morbidity and mortality associated with EBR, its use is restricted to certain types of tumor. To decide whether a tumor is appropriate for EBR, its behavior and characteristics must be taken into consideration. This is done by applying the Enneking classification.[2] This is a type of oncological staging that was first developed in reference to tumors of the long bones but has since been applied to the spine. (►**Table 2**)

Benign Tumors

The Enneking staging system is split into benign and malignant mesenchymal tumors. The benign tumors are split into three categories depending upon the radiological appearance of their borders. Well-demarcated borders represent more latent lesions, whereas indistinct borders reflect more aggressive lesions.[5] The rate of recurrence increases with an increase in grade. Metastasis from locally aggressive benign lesions is rare but can occur in giant cell tumors (GCTs) and chondroblastoma.[6] EBR is felt appropriate in cases of benign aggressive (Enneking stage 3) tumors such as GCTs, chondroblastomas, and osteoblastomas.[5]

Table 2 Enneking staging system

Stage 1
Low-grade intracompartmental (A) Low-grade extracompartmental (B)
Stage 2
High-grade intracompartmental (A) High-grade extracompartmental (B)
Stage 3
Intracompartmental with metastasis (A) Extracompartmental with metastasis (B)

Giant Cell Tumor

GCTs of the spine are most commonly found in the sacral region and are more common in women and patients aged between 20 and 40 years.[7,8] In the mobile spine, GCTs most often occur in the thoracic segment, followed by the lumbar and cervical region.[9] They are usually discovered at Enneking stage 2 or 3[9] and the reported recurrence rate is high (11–45%).[10] About 13.7% of GCTs of the spine will eventually metastasize to the lung, a higher rate than GCTs of the extremities.[11] A recent meta-analysis of studies comparing EBR with intralesional resection (IR) of spinal GCT found that the local recurrence rates and rates of postoperative complications were decreased with EBR. Local recurrence rates were 36.7 and 9.5% in the IR and EBR groups, respectively. Rates of postoperative complications were 36.4% with IR and 11.1% with EBR.[12] (Obviously, these results will be subject to a selection bias by the operating surgeon as it is likely that surgeons would prefer EBR in younger patients with fewer comorbidities and less tumor extension). It is important to note that although EBR is associated with better local control of Enneking stage 3 GCTs, IR provides adequate control of Enneking stage 2 tumors.[8] This highlights the importance of reviewing the radiological appearance of the borders and extent of the lesion to determine the Enneking stage.

Osteoblastoma

Forty percent of osteoblastoma lesions (OBL) occur within the spine,[13] particularly the cervical and lumbar regions.[14] Within the vertebra, OBL are typically found in the posterior elements, mostly the pedicle or lamina.[15] OBL typically occur in the second decade of life[16] with ~80% of patients presenting before the age of 30.[15] Similarly to GCT, OBL can present at Enneking stage 2 or 3. Enneking stage 3 OBL, sometimes described as “aggressive” variant, are associated with a 50% recurrence rate after IR, stage 2 OBL or “less aggressive” variant are associated with a recurrence rate between 10 and

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15%.[17] For this reason, IR is recommended for “active” OBL (Enneking stage 2), while EBR, when anatomically feasible, is recommended for “aggressive” OBL (Enneking stage 3), again highlighting the importance of an analysis of the radiological appearance of the borders of the lesion.

Malignant Tumors

The staging of malignant tumors considers surgical grade, local extent, and presence of metastasis. Low surgical grade lesions have a low risk of distant spread with low mitotic rates, low nuclear to cytoplasmic ratio, and limited pleomorphism. High-grade lesions have a higher incidence of metastasis and are characterized by mitotic figures, prominent nucleoli, and pleomorphism.[5] Most lesions are graded based on their histology; however, some tumors fall into the high-grade category by definition, for example, dedifferentiated chondrosarcoma.[18] Radiology plays an important role not only in assessing the position of the tumor (intra- or extracompartmental) but also in describing the presence of regional or distant metastasis. EBR is felt appropriate in low-grade malignant tumors (Enneking stage 1A and 2B) such as chordomas and chondrosarcomas. High-grade malignancies (Enneking stage 2) require consideration of chemotherapy and radiotherapy (RT). The role of EBR in spine metastasis (Enneking stage 3) is very limited and only considered in selected cases if the multidisciplinary team feel it would improve local control and reduce local recurrence. This will not be discussed in this article.

Chordoma

Chordomas are rare bone tumors derived from embryonic notochord remnants that mostly involve the skull base, sacrum, and spine.[19] They are locally aggressive tumors with the ability to metastasize and can often recur locally.[20] They occur twice as often in male compared with female patients and are most likely to present in the sixth decade.[19] Total EBR with a circumferential margin of uninvolved tissue was initially considered the only treatment option associated with disease-free survival at 5 years follow-up.[21] Recently, proton beam radiation therapy has become first line in the treatment of spinal chordoma as it has proven to decrease local recurrence even further following maximal resection.[22]

Chondrosarcoma

Chondrosarcoma is a rare, low-grade malignant chondroid lesion that occurs in the spine in less than 10% of cases.[23] They can occur at any level in the mobile spine but are more commonly seen in the thoracic and lumbar regions.[24] The average age at diagnosis is 42 years, with men more commonly affected than women.[25] The tumor is renowned for being difficult to treat with radiation and chemotherapy.[26] The extent of surgical resection is correlated with the overall survival benefit with EBR being the gold standard of treatment.[27] The reduced relative risk (RR) comparing en bloc with other surgical techniques for recurrence and mortality is 78.8% (RR, 0.21; $p < 0.001$) and 80.7% (RR, 0.19; $p < 0.001$), respectively.[25,26,28]

Osteosarcoma

Although osteosarcoma is one of the more common primary bone malignancies, it most often affects the extremities with only 0.85 to 3% of all osteosarcomas affecting the spine.[29] It presents later than extremity osteosarcoma around the fourth decade[30] and is most commonly seen in the thoracolumbar spine, especially the posterior elements.[31] Although adjuvant chemotherapy and RT play important roles in the management of spinal osteosarcoma, surgical resection of the lesion is critical[27] Improved long-term prognosis and decreased local recurrence following total en bloc with wide or marginal margins have been demonstrated throughout the spine, compared with intralesional or “piecemeal” resection.[27,32]

Ewing Sarcoma

After osteosarcoma, Ewing sarcoma (ES) is the second most common primary malignant bone cancer in children and adolescents.[33] It most often affects the pelvis, femur, and tibia, and the spine is only affected in 5% of cases.[34] When the spine is involved, ES tends to be seen in the thoracolumbar spine and originates in the posterior elements with extension into the vertebral body.[35] The prognosis is worse in the spine and there has been debate regarding the optimal method of local control. RT has traditionally been considered the primary component of local therapy; however, this is limited due to the proximity of the spinal cord and the kidneys in lumbar spinal tumors.[36] A limited number of small studies consider the effect of total enbloc resection (TER) on local recurrence and disease-free survival. Combining these small studies, it has been shown that where possible TER combined with RT compared with RT alone provides superior local control.[37] IR or EBR with IL margins demonstrate worse local control than RT and chemotherapy combined.[38]

Standardization of Approach (WBB Staging) Surgical Staging

The WBB surgical staging system was introduced in 1997 with the aim of assisting and standardizing the planning of EBR.[1] It has since been subject to clinical evaluation and is the system used in most spinal oncology studies and has been adopted by most spinal oncology centers. This staging system categorizes tumors based on their location in terms of zones and layers and the surgical approach is subsequently determined by this.[2] A total of 10 different surgical strategies are described.[2] It would be beyond the scope of this article to give an in-depth explanation of all 10, but the radiologist should be aware of the three main approaches to help tailor their reports. The three main approaches are as follows.

Vertebrectomy (Marginal/Wide En Bloc Excision of the Vertebral Body)

This approach is suitable for tumors located centrally in the vertebral body with at least one pedicle spared (confined to zones 4–8 or 5–9). It involves initially removing the healthy posterior elements via a posterior approach. This exposes the annulus fibrosis and posterior longitudinal ligament and

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enables them to be sectioned. A subsequent anterior approach then allows ligature of the segmental vessels at the operative level above and below. The vertebral body is then removed en bloc. This is done either via proximal and distal discectomies or by sectioning through the neighboring vertebral bodies. It is therefore important for the radiologist to accurately describe the longitudinal extent of the tumor and provide detail as to whether there is involvement of the adjacent intervertebral disc. This technique removes the entirety of the vertebra including the body and posterior elements.

Sagittal Resection (Marginal/Wide)

This is the most appropriate approach for tumors, which are located eccentrically in the body of the vertebra or within the pedicle or transverse process (confined to zones 3–5 or 8–10). More than one level can be removed, as can one or more of the ribs if necessary. The first step is via a posterior approach and the healthy posterior elements are removed, including the unaffected pedicle. Next, the nerve roots at the affected levels are ligated. The patient is then moved into a lateral decubitus position and the eccentric tumor in the vertebral body or transverse elements is approached anteriorly. A chisel or osteotome is used to cut through the vertebral body through at least one zone distant to the tumor. The diseased elements are then removed en bloc. This approach will leave a small section of healthy vertebral body.

Resection of the Posterior Arch (Marginal/ Wide)

When the tumor is confined to the posterior elements alone (zones 10–3), it can be removed en bloc via the posterior approach alone. Initially a wide laminectomy is performed at the vertebral level above and below the tumor to expose the dural sac. The posterior elements at the affected level are then removed en bloc by sectioning the pedicles with an osteotome or Gigli saw.

The Radiological Report

Having considered the surgical options, the following is a suggested approach to reporting preoperative planning imaging for EBR.

Longitudinal Extent of the Tumor

The vertebral level at which the epicenter of the tumor lies should be given. Particular consideration of transitional vertebra should be considered and it should always be clear in the report is the radiologist is counting the vertebra from the sacrum up or from C2 down to avoid wrong level surgery. The report should also detail the longitudinal spread of the tumor. An important surgical decision is to where to make the longitudinal margins. If the tumor is invading the disc above or below, the surgeon will resect through the vertebral body above. If the disc is spared then the affected vertebral body can be resected by performing superior and inferior discectomies.

Transverse Extent of the Tumor

To allow application of the WBB staging system and to assist the planning of approach, the radiologist will need to accurately describe the location and extent of the tumor in the transverse plane. This can be done by describing the location of the tumor in terms of the WBB zones and layers. The vertebrae are divided into twelve radiating zones (1–12) in a clockwise manner and into five layers (A-E), which can be used to describe the position and extent of the tumor. When a tumor extends into layer A (extra osseous soft tissue), it is important to describe what structures that tumor is invading or abutting, for example, any retroperitoneal structures in the lumbar spine, or mediastinum, pleura, or lung at the thoracic level (►**Fig. 2**). The osseous involvement needs to described in particular if there is superficial or deep intraosseous component (►**Fig. 1**). This might limit the resection margins or prompt the involvement of additional surgical specialities during definitive surgery.

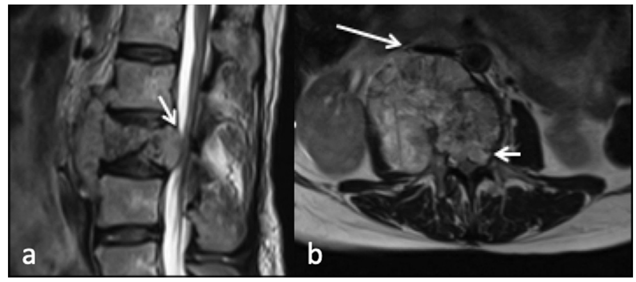

Fig. 2 T2 sagittal (A) and axial (B) showing large chordoma with epidural component (small arrow). Tumor is compressing but not encasing the inferior vena cava (long arrow) (Weinstein-Boriani-Biagini 4–10, **a–d**).

Involvement of Neural Structures

A key feature of importance for the spinal surgeon is knowing whether there is an extension of the tumor into the vertebral canal. Using the WBB staging system, this can be described as extending into layer D (extra-osseous extra-dural) or layer E (extraosseous intradural). While it may be possible to peel an extraosseous extradural tumor away from the spinal cord and therefore achieve adequate margins, obviously, this will be limited in an extraosseous intradural tumor (►**Fig. 2**).

When considering resection of a spinal tumor with the same oncological principles as resection of a mass in the long bones, any invasion of the tumor into the vertebral canal will mean contamination of that compartment. Clearly, the entire compartment cannot be removed, and therefore patients should be warned about any implications this may have on prognosis and the need for adjuvant therapy. In extreme cases, where there is a significant invasion of neurological structures, these may need to be sacrificed. There have even been reports of the sacrifice of the cauda equina.[38] Clearly this is not a decision made lightly and the benefits and limitations need to be discussed with the wider multidisciplinary team and the patient.

Separate to the cord itself, it is also important to describe any invasion or encasement of the segmental nerves by the tumor mass. Again, the surgeon will need to balance the functional loss caused by sectioning these nerves with the benefits of achieving clear oncological margins.

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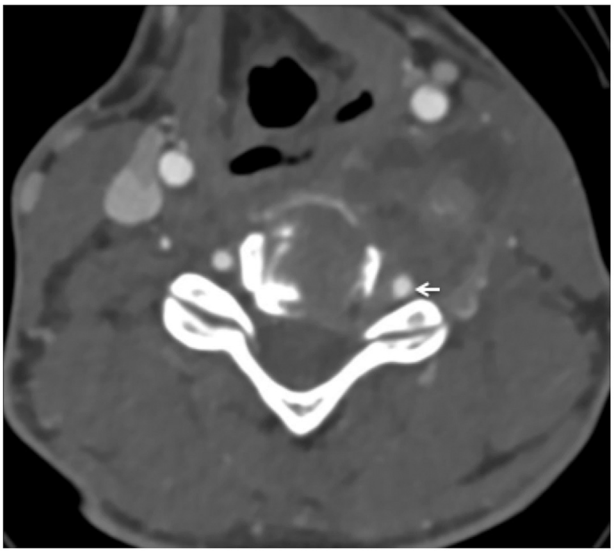

Fig. 3 Axial computed tomography angiogram image showing tumor displaced left carotid vessels anteriorly and encasing left vertebral artery (arrow).

Involvement of Vascular Structures

It is important, where possible, to describe tumor invasion or encasement of any surrounding vascular structures (►**Fig. 3**). The most obvious would be any involvement of the aorta or vena cava, which in most cases may prove EBR an unfeasible treatment option. (►**Figs. 2** and **4**). There has been a case report of a successful combined surgery that involved EBR of an osteosarcoma with the replacement of an involved section of the aorta.[39] Therefore, when reporting vascular involvement, the extent and length of the involved area are important to describe. The lumbar vertebrae pose particular challenges due to their proximity to the arterial (aorta) and venous (IVC) bifurcations. For this reason, dedicated surgical approaches are described for lumbar tumors and tumors at L5.[2]

Within the cervical spine, a specific concern is of involvement of the vertebral artery. Of course, it is important to describe any tumor invasion and to report whether there is extensive encasement of the vessel, which may make it difficult to resect a lesion en bloc. Again, this may prevent EBR entirely, but there may be ways to overcome this. There have been cases describing unilateral ligation of a vertebral artery to allow successful en bloc removal of a lesion without neurological compromise. In such cases, it would be advised to perform a formal computed tomography (CT) angiography to assess for variations within the circle of Willis and to perform an occlusion test (►**Fig. 3**).

Assessment of the smaller segmental/ radicular arteries is unlikely to be possible on CT or magnetic resonance imaging (MRI) due to their small size and possible disruption due to the surrounding lesion. In several of the approaches described above, ligation of the segmental artery at the affected level and at the level above and below the lesion is necessary. It may also be necessary to embolize these vessels preoperatively when the tumor is predicted to be highly vascular in an attempt to decrease intraoperative blood loss or the risk of massive hemorrhage.

Initially, in these cases, angiography was used to determine the relationship between the segmental vessels and the spinal arteries especially the artery of Adamkewitz. However, it has since been demonstrated that even when ligating the main segmental artery feeding into the artery of Adamkewitz, neurological defects are highly unlikely. This is considered to be due to collateral supply to the anterior spinal artery. What is felt to be of greater importance is the number of segmental arteries that are cut, with the risk of neurological problems rising significantly when more than three pairs of segmental vessels are cut. If the surgeon feels that preoperatively this may be necessary, then angiography could be useful in these cases. An initial small study has assessed the role of electrophysiological monitoring during preoperative embolization of major radicular arteries in guiding decisions regarding permanent occlusion of these vessels, which may prove useful in particularly challenging cases.[40]

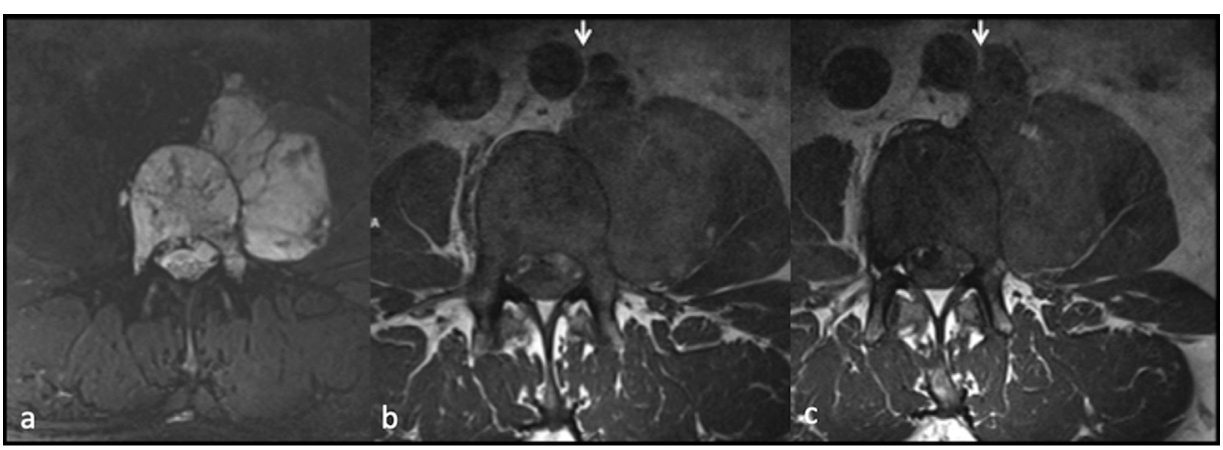

Fig. 4 Axial short tau inversion recovery (**A**), T1 (**B** and **C**) showing large chordoma that is closely related to the aorta with loss of fat plane between the tumor and aorta on image (**C**) (arrow). The tumor extends and involves the left psoas. (Weinstein-Boriani-Biagini 3–10, **a–d**).

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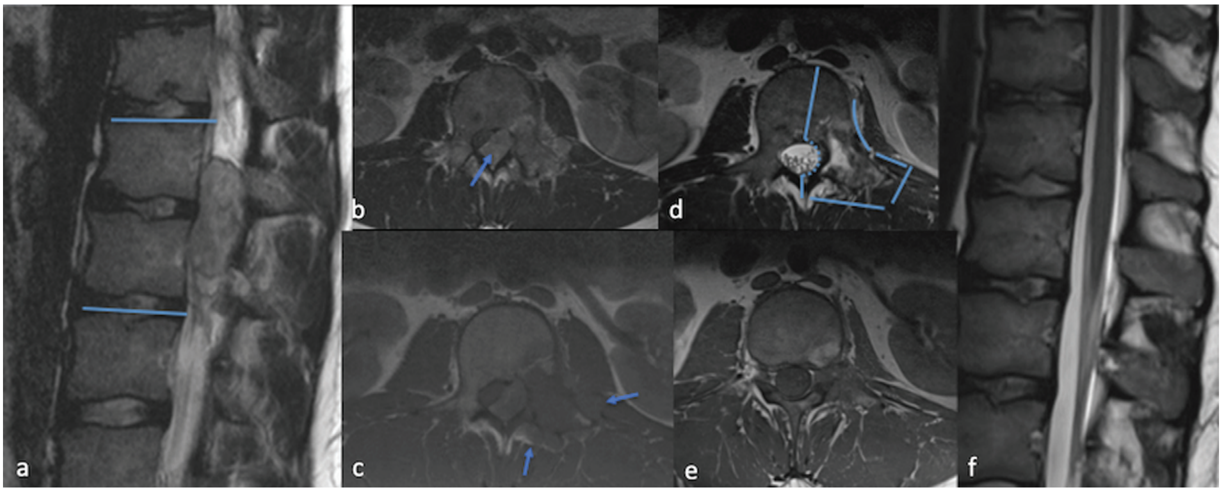

Fig. 5 Ewing's sarcoma. Sagittal T2 (**A**), axial T2 (**B**), T1 (**C**), post chemotherapy axial T2 (**D**), T1 (**E**), and sagittal T2 (**F**) showing tumors in the canal and extending into the left psoas (arrow). (Weinstein-Boriani-Biagini 2–5, **A–E**). The tumor has decreased significantly post chemotherapy and blue lines denote the resection margin.

Stability of the Vertebra

This will be best assessed on the preoperative CT imaging. A comment should be made regarding the extent of bony invasion, especially the breakdown of cortical structures. From this, deductions can be made about the bony stability of the vertebra involved and also the vertebral levels above and below. This is of particular importance to allow reconstruction following the EBR.

Cases

►**Figs. 5**,**6**,**7**,**8**.

Conclusion

EBR of vertebral tumors is a complex and demanding surgery with the potential to greatly improve disease-free survival and limit local recurrence. When considering the appropriateness of EBR, the Enneking staging system is used to predict the behavior of the lesion in question. Following this, the WBB staging system is used to help plan the surgical approach. When reporting preoperative planning for EBR, the radiologist should focus on five major domains including longitudinal extent of the tumor, the transverse extent of the tumor, involvement of neurological structures, involvement of vascular structures, and the stability of the vertebra (►**Table 3**). This will assist the surgeon in planning the surgical approach and help to predict any intraoperative complications. Preoperative MRI and CT can also aid to create three-dimensional model to aid in surgical planning and navigation.

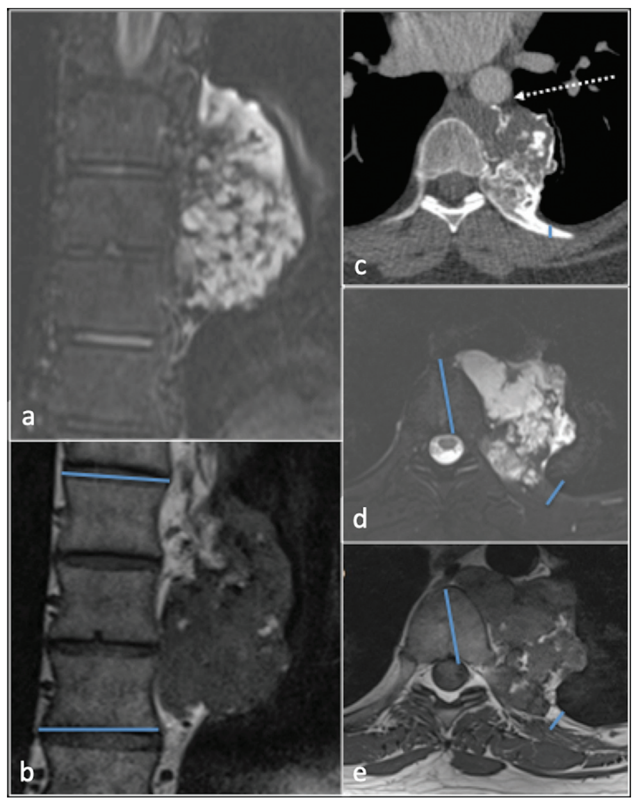

Fig. 6 Chondrosarcoma. Coronal short tau inversion recovery (STIR) (**A**) T1(**B**), axial computed tomography (**C**), axial STIR (**D**) and T1 (**E**) showing resection margins (blue lines). Arrow showing the tumor is close to the aorta. (Weinstein-Boriani-Biagini 2–6, **A**).

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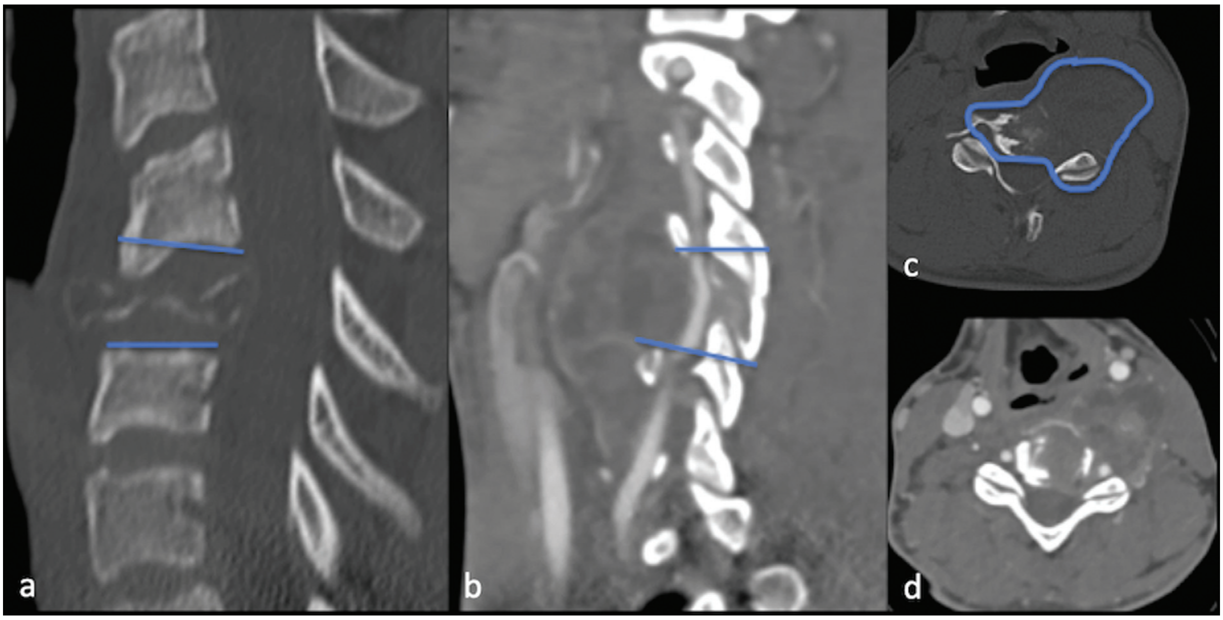

Fig. 7 Aneurysmal bone cyst. Sagittal computed tomography (CT) (bone windows) (**A**), sagittal CT angiogram (**B**), axial CT angiogram (bone window (**C**), and soft tissue window (**D**) showing site of resection (blue lines). The left vertebral artery and left C6 nerve root need to be resected. (Weinstein-Boriani-Biagini 2–9, **a–c**).

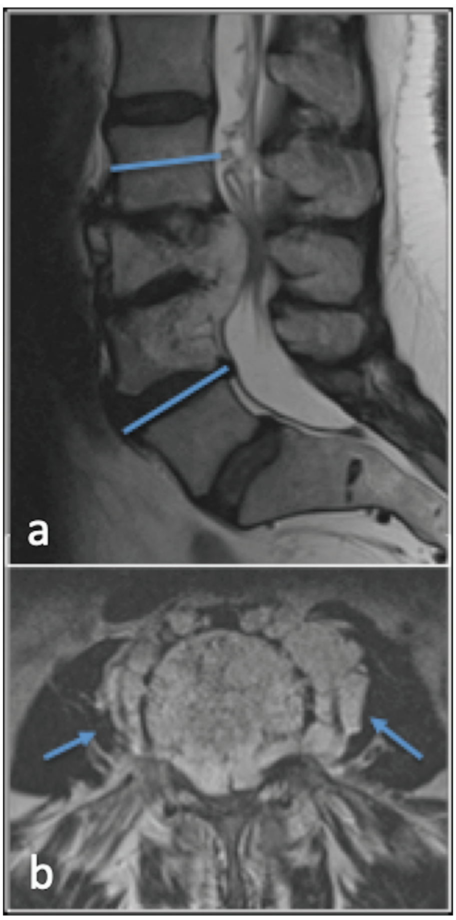

Fig. 8 Chordoma. Sagittal T2 (**A**) and axial T2 (**B**) showing resection marking (blue lines). The arrows demonstrate that the psoas muscle is closely related to the tumor and might have to be resected. (Weinstein-Boriani-Biagini 4–9, **a–d**).

Table 3 Reporting template

Template
1. Longitudinal extent of tumor
2. Transverse extent of tumor as per WBB
3. Stability—extent of bony destruction (best assessed on CT)
4. Neural structure involvement—cord and nerve roots
5. Vascular structures, in particular vertebral artery, segmental artery, aorta, inferior vena cava

Abbreviations: CT, computed tomography; WBB, Weinstein-Boriani-Biagini.

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Conflict of Interest
No conflicts of interest.

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