



***En bloc* resection in the spine: a procedure of surgical oncology**

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Provenance: This is an invited Editorial commissioned by Section Editor Dr. Ai-Min Wu (Department of Orthopaedics, The Second Affiliated Hospital and Yuying Children's Hospital of Wenzhou Medical University; Key Laboratory of Orthopaedics of Zhejiang Province, The Second School of Medicine, Wenzhou Medical University, Wenzhou, China).

Comment on: Shah AA, Paulino Pereira NR, Pedlow FX, *et al.* Modified En Bloc Spondylectomy for Tumors of the Thoracic and Lumbar Spine: Surgical Technique and Outcomes. J Bone Joint Surg Am 2017;99:1476-84.

Submitted Apr 10, 2018. Accepted for publication Aug 02, 2018.

doi: 10.21037/jss.2018.09.02

View this article at: <http://dx.doi.org/10.21037/jss.2018.09.02>

It is really stating the obvious to say that a surgical procedure aiming to remove a tumor should be planned based on oncological principles.

En bloc resection is a procedure of surgical oncology aiming to remove a tumoral mass in its entirety, completely surrounded by a continuous layer of healthy tissue

The healthy tissue surrounding the tumor has been named “margin”: its quality and its thickness qualifies the procedure from an oncological point of view, affecting the local and systemic prognosis (1,2). This procedure became the golden standard in the treatment of bone tumors of the limbs in the seventies, after the introduction of the protocols of neo-adjuvant chemotherapy. The effects of these new drugs on the tumoral mass (volume reduction, harder consistency) allowed to develop techniques of surgical resection the tumor without sacrificing the limb (so called “limb salvage procedures”) (3).

En bloc resection of spine tumors obviously require a deep knowledge of specific regional issues of surgical anatomy, as the margins are possibly represented by anatomical structures of relevant functional role.

Reading the literature on spine tumors, the oncological principles seem to receive less consideration than the details of surgical techniques and the application of up-to-date technologies (4).

En bloc resection in spine tumors requires spine surgery skill and multidisciplinary competences, and is a very interesting argument for discussion and sharing different opinion and experiences: newest techniques should be applied to improve the outcome and to make surgery less

difficult, less morbid and more reproducible, but the use of these new tools should always be secondary to the fulfilling of oncological principles. Technologies are the means, not the end.

Some papers are dedicated to functional results (5) undeniably important, but—differently from metastases—secondary to oncological principles in the decision-making process of primary tumors

Focus should be concentrated on the local recurrence rate, which is the best indicator of the validity of a procedure of surgical oncology (2).

Surgical techniques to perform *en bloc* resection in the spine have been frequently proposed irrespective of tumor extension: the most popular technique of *en bloc* resection of a spine tumor, described by Roy-Camille *et al.* (6) and later by Tomita *et al.* (7) has an oncological validity only if the tumor is not growing over the antero-lateral vertebral body cortex, otherwise the blunt manual dissection will breach the tumor margin.

Tomita proposed the term Total *En bloc* Spondylectomy, which in my perspective, is not oncologically appropriate. In fact, the target is not to resect *en bloc* the whole vertebra, but to resect *en bloc* the tumor with an appropriate margin. This sometimes does not require to remove the whole vertebra.

Bertil Stener was the pioneer of the application to the spine the oncologic principles generally accepted for the gastrointestinal tumors (8). His detailed reports of the surgical planning of *en bloc* resections are till now extremely useful and exhaustive as a guide to learn how

oncological principles can guide the surgical planning. His work represents a watershed between precedent pioneers' activity and strict adherence to the oncological principles at that time developed. It is here the place to acknowledge the experiences by Janos Szava, professor of Orthopedics at Marosvásárhely (Romania). He performed some spine tumor resections in the 1950s which are still unknown by most of us due to language and political barriers (9).

Several articles were later on written describing different approaches and different combinations of approaches. Some of them are particularly relevant as have a strong oncological commitment, targeting the surgical procedure on the achievement of a full free-tumor margin resection even with the sacrifice of relevant anatomical structures: dura (10), cervical nerve roots (11) cauda equina and spinal cord (12), major vascular structures and visceral organs (13).

Among the details of technique, the technique of osteotomy is the most discussed one. Roy-Camille (6) and Stener (8) years before, proposed to perform the osteotomy by the combined use of a Gigli and osteotomes. In those papers it is stressed on the risk of losing the control of the Gigli saw during the final steps ending in incidental injury of the dural sac. Tomita (7) proposed a thinner saw and an original set of instruments—relying on the hands of the assistant—both to protect the dura from accidental injuries while cutting anterior to posterior, both to perform a coronal section of the pedicles, allowing to finalize the spondylectomy by achieving two specimens.

An original proposal by Gasbarrini *et al.* is the malleable protector of the dura (14) to be inserted between the dura and the posterior vertebral wall and fixed to one of the rods: it is a solid and sound protection from accidental injuries without relying on the hands of a surgeon.

In a recent well documented article by Shah *et al.* (15) an interesting use of the threadwire saw is proposed. It is an interesting tip, whose application however is limited to some part of the thoracic and lumbar spine: it is not applicable to high thoracic spine neither to low lumbar spine.

According to different personal experiences and manuality, chisels, osteotomes, ultrasound osteotome, high speed burr can be indifferently used to perform the osteotomies without affecting the final outcome, provided the resection is finally achieved with appropriate margin. For the purpose of a sound and balanced reconstruction, a perfectly flat osteotomy surface should be obtained, for a full contact with the cage and/or the graft.

It should also be considered that according to the surgical planning, discectomy (and all the relative tools) can

be preferred to osteotomy. In this case, all disk material and cartilage should be removed from the endplates for a better cage positioning and graft incorporation.

In my opinion, we should follow the great message delivered by Bertil Stener in his unforgotten papers: it does not exist a single surgical technique able to perform *en bloc* resection of all bone tumors in the spine, but the surgical technique should be planned according to the tumor extension, the spine location, the histology, the margins to be achieved (8).

The new frontier is to consider the possibilities offered by new technologies of radiation therapy (RT) and new protocols of chemotherapy to recover margin transgression incidentally occurring or intentionally decided to save anatomical structures according to the patient preferences.

Oncological basis for treatment

The Enneking staging system (1) is a valid and reproducible tool for understanding and staging the biological behavior of bone and soft tissue tumors and for deciding the appropriate surgical procedure from an oncological point of view. This system is based on histological diagnosis and on clinical, laboratory and imaging studies. It also proposed a common terminology to the multidisciplinary team who take care of these diseases.

For simplicity purpose the surgical procedures following the dictates of the Enneking staging system are defined as Enneking appropriate (2)

En bloc resection is recommended in cases of benign aggressive (Enneking stage 3) tumors (i.e., osteoblastomas and giant cell tumors) and low-grade malignant tumors (Enneking stage I A and B) like chordomas and chondrosarcomas. In high grade malignancies (Enneking stage II) like osteosarcoma and Ewing sarcoma chemotherapy and radiotherapy have a very relevant and essential role.

Once the resection performed, the pathologist must carefully evaluate the tumor margins, as defined by “wide” (a relevant barrier like a fascia or at least healthy bone 1 cm thick) “marginal” (a thin barrier like periosteum) or “intralesional”.

“Intralesional” resection is defined when the surgeon incidentally or intentionally violates the tumor. Violation of the margins significantly worsen the prognosis (2). Intentional intralesional resection [so called intentional transgression to oncological principles (2)] may be an option when the patient does not accept the sacrifice of a

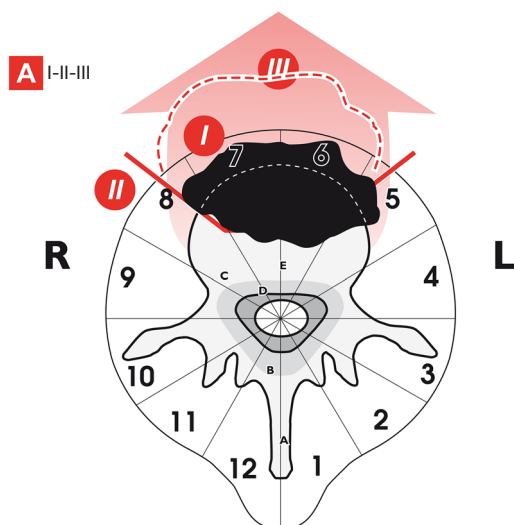


Figure 1 Type 1 WBB-based *en bloc* resection. Single anterior approach. For achieving a tumor-free margin the tumor must be only in sectors 8 to 5, layer A and B, but not layer C. In this case in fact, the osteotomy would violate the tumor mass: a posterior approach is necessary for a tumor free margin resection by including the posterior wall in the resection by entering the canal and releasing the dura. Three steps are to be considered: the first to provide under direct visual control an appropriate margin of the anterior tumor growing (I). The second step include performing an osteotomy between the tumor and the posterior wall (II). The third step is the tumor removal (III). WBB, Weinstein-Boriani-Biagini.

functionally relevant element that is closely contiguous to the tumor or has been infiltrated.

The patient however must be fully informed of the higher risk of recurrence after Enneking-inappropriate procedure (2) and that the rate of complications and further tumor recurrence are significantly higher after revision surgery (16).

If patient strongly requires the preservation of anatomical structure to save the function, and notwithstanding the exposition to higher recurrence rate, adjuvant therapy is indicated.

En bloc resection has a limited role in the treatment of spine metastases. The primary goal in these patients is to preserve or improve function and quality of life without unnecessary morbidity. Giving the priority to function, no major anatomical sacrifice with consequent relevant loss of function should be planned. However, in some selected cases, after a multidisciplinary discussion *en bloc* resection could be proposed to reduce or delete any risk of local

recurrence. In the authors' experience, the indication to *en bloc* resection is appropriate in single localizations, with full tumor control at the primary site and no involvement of visceral organs, best after long term disease free evolution. The key point in this decision is the lack of sensitivity to medical oncology or radiation oncology treatments: alternatively, less aggressive surgery could be combined with these treatments, reducing the surgical morbidity without reducing the possibility to local cure.

Surgical planning

The Weinstein-Boriani-Biagini (WBB) surgical system was proposed in 1997 (17) to stage the extension of spine tumors. It has been adopted in several spine oncology centers and is used in most of spine tumors related articles. The WBB system has been submitted by an international multidisciplinary group of spine tumor experts (18) to a reliability and validity study resulting in a moderate interobserver reliability and substantial intraobserver reliability.

The WBB staging system (17) can be helpful in surgical planning of *en bloc* resection.

Accordingly, seven types of procedures are here proposed, defined by the approach or the combination of approaches, with several subgroups, ending in a total of ten different surgical strategies.

Single anterior approach (type 1); single posterior approach (type 2) including three subtypes (a, b, c); anterior and then posterior approach (type 3) with three subtypes (a, b, c); first posterior approach, followed by both side anterior approaches (type 4); first posterior approach and then simultaneous anterior and reopening of posterior approach (type 5); anterior, posterior, and then simultaneous anterior (contralateral) and reopening of posterior approach (type 6, mostly performed for L5); posterior approach first and anterior approach as second step (type 7).

Type 1: Single anterior approach (Figure 1) allows to perform *en bloc* resection only of small volume tumors arising in the vertebral body of the thoracic and lumbar spine.

Type 2: Single posterior approach allows to perform many different *en bloc* resections either on tumors occurring in the posterior elements, either in the vertebral body either eccentrically located (Figure 2).

Type 3: Anterior approach first, posterior second is the strategy proposed to perform *en bloc* resection of cervical spine tumors (Figure 3A) which involve part of the vertebral

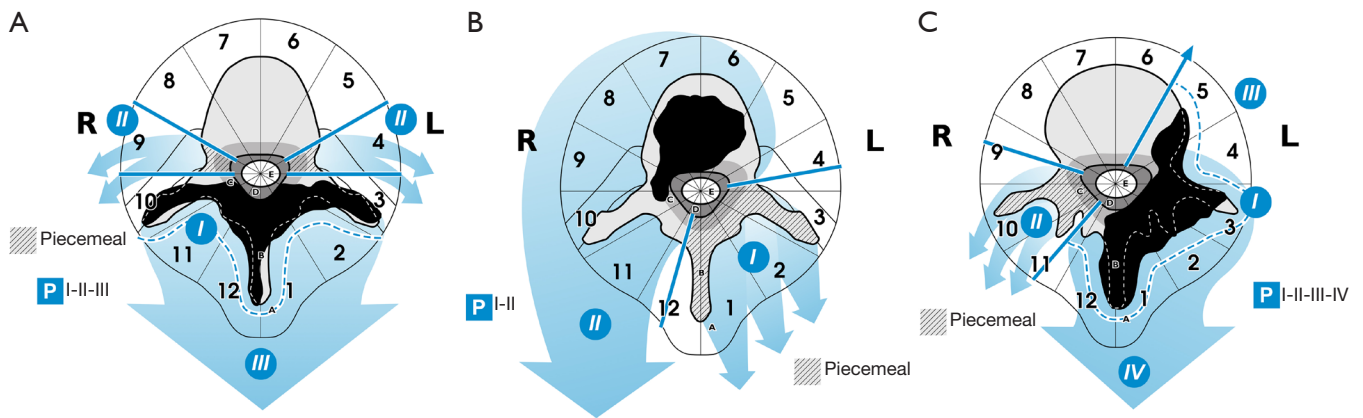


Figure 2 Type 2 WBB-based *en bloc* resection. (A) Type 2a WBB-based *en bloc* resection. Single posterior approach, *en bloc* resection of a tumor arising in the posterior arch. To achieve an appropriate margins sector 9 and 4 must be free from tumor. If the tumor grows in layer D, a margin violation will result intralesional during the dura release. This technique includes three steps: first to leave an appropriate margin over the posterior tumor mass (I). Second step to enter the canal by excision of sectors 9 and 4 (II). The third step is a transverse laminotomy above and below, the tumor release from the dura and the *en bloc* tumor removal (III). (B) Type 2b WBB-based *en bloc* resection. Single posterior approach. It allows to remove by *en bloc* resection a thoracic spine tumor growing in the vertebral body. Criteria suggested for a tumor free margins are that sector 9 or 4 be not involved by tumor. If the layer D is involved by the tumor, the margin can be violated intralesional during the release from the dura. If the tumor extends in layer A, the margin can be violated during the separation from the anterior structures. This is the most popular technique of *en bloc* resection of a spine tumor, as described by Roy-Camille *et al.* (6) and later by Tomita *et al.* (7). The digital blunt release of the anterior spine from the mediastinum should be performed before laminectomy to reduce the risk to damage the cord during the manual dissection. The aorta is safer released from the anterior spine wall if the segmental vessels are identified and the blunt dissection performed between the segmental vessels and the vertebra. Uninvolved posterior arch excision is then performed. At least 4 sectors should be removed, starting from sector 4 or from sector 9 (I). Section after ligation of the nerve root(s) involved by the tumor is necessary and dura must be fully released from the tumor. Discectomy or osteotomy is then performed above and below the tumor, and the tumor removal is completed (II). (C) Type 2c WBB-based *en bloc* resection. Single posterior approach to resect eccentrically growing tumors in the thoracic or lumbar spine with sagittal osteotomy. Criteria to achieve *en bloc* tumor free margin resection are the vertebral body not involved over sector 5 at left and over sector 8 at right and at least 3 posterior sectors not involved (4 to 1-2 or 12-11 to 9). This technique includes four steps: the first to provide a tumor free margin over the posterior growing tumor (I). The release should proceed laterally till the lateral side of the vertebral body. In the thoracic spine the pleura can be left on the tumor, in the lumbar spine the posterior part of the psoas must be dissected, but the segmental vessels must be found and ligated. The step II is the excision of the posterior arch not involved by the tumor to approach into the canal; the dura is released from the tumor (if the tumor grows in layer D, the margin can result intralesional) and the nerve root(s) involved by the tumor are sacrificed. In step III the dura is carefully displaced and osteotomy is performed from posterior to anterior in sector 8 or 5. Step IV is the tumor removal. WBB, Weinstein-Boriani-Biagini.

body (no sector 6 and 7, otherwise type 4 is suggested) and part of the posterior arch (at least 3 sectors should be not involved) or in tumors located in the thoracic and in the lumbar spine when the tumor is growing anteriorly in layer A (Figure 3B), or in case of tumor eccentrically growing in the thoracic and lumbar spine (Figure 3C,D) when sagittal osteotomy is considered safe for appropriate margin, without need to remove the whole vertebral body.

Type 4: In some huge tumors of the cervical spine, extending over the midline, three approaches are required for a safe and oncologically appropriate surgery: first step

is a posterior approach, the second step is an anterior approach contralateral to the tumor, the third step is an anterior approach on the tumor side (Figure 4)

Type 5: This includes two stages: first a posterior approach, then a combined anterior and posterior approach with the patient positioned on side (Figure 5). This demanding technique (associated with the highest rate of morbidity and complications) can be the most appropriate for lumbar tumors expanding anteriorly. This technique was described by Roy Camille for lumbar tumors (6) and is associated with the highest rate of morbidity and

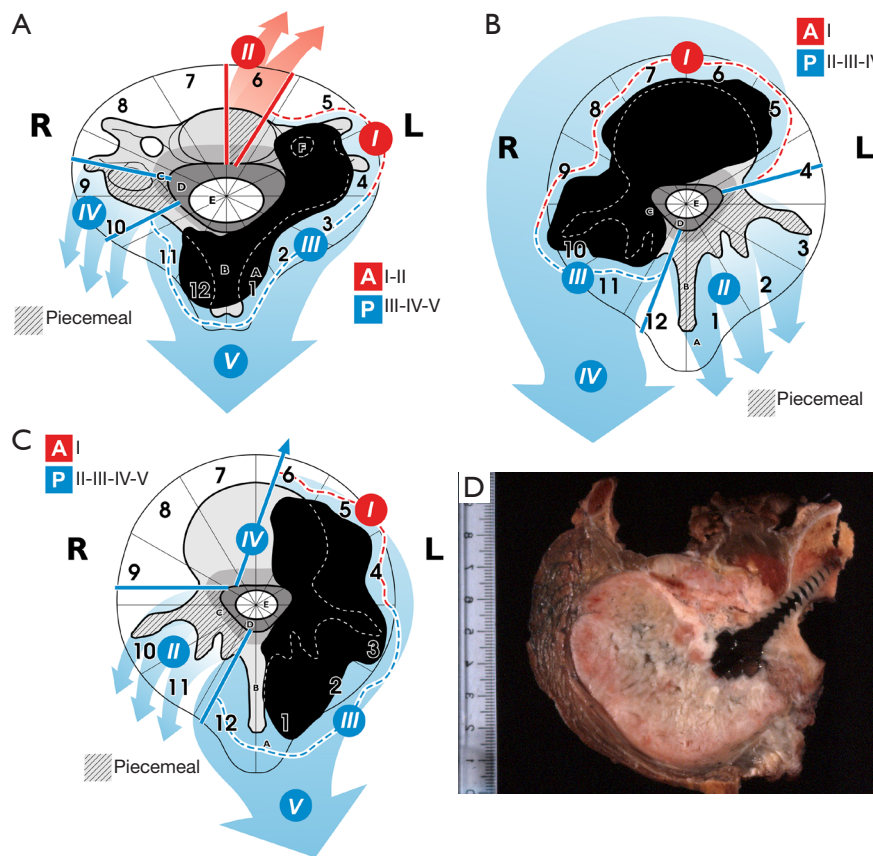


Figure 3 Type 3 WBB-based *en bloc* resection. (A) Type 3a WBB-based *en bloc* resection. This technique is specific for some cervical spine tumors and requires three approaches: first posterior, second anterior contralateral to the tumor side, third anterior on the tumor side. A single wide transverse anterior approach can be considered. The first step is a common posterior approach in prone position and is finalized to resect the posterior arch not involved by the tumor. At least 3 sectors are needed, from sector 4 or from sector 9 (I). If the tumor has a posterior extension in layer A, a margin must be provided by resecting inside the posterior muscles covering the tumor mass (II). The dura must be released from the nerve root(s) crossing the tumor sacrificed. The second and third steps are performed in supine position. In step II a sagittal groove is performed in the vertebral body not occupied by the tumor and the vertebral artery isolated and protected as the other is involved by the tumor and must be sacrificed. Step III: the upper and lower margins are defined by diskectomies or transversal grooves in vertebral bodies as planned, including ligation of the vertebral artery if necessary. The tumor is finally removed. (B) Type 3b WBB-based *en bloc* resection: when a thoracic or lumbar tumor is growing anteriorly (layer A) an anterior approach must be performed as first step to provide a wide/marginal margin under visual control. In case of tumors mostly occupying the vertebral body, the anterior approach can be the first step to release from mediastinum or retroperitoneal, eventually leaving involved structures as margin (I). A sheet of silastic or similar can be left as protection. Second stage, posterior approach: piecemeal excision of the posterior arch not involved by the tumor (II). At least 3–4 sectors are required, starting from sector 4 or from sector 9. Release of the dura from the tumor, section of the nerve root(s) involved by the tumor, then provide the appropriate margin over the tumor posteriorly growing by resecting inside the posterior muscles covering the tumor mass if it is expanding in layer A (III). Finally, the specimen is removed by rotating around the dural sac (IV). (C) Type 3c WBB-based *en bloc* resection. When a tumor is arising eccentrically and growing anteriorly (layer A) if and appropriate margin can be left, a sagittal or oblique osteotomy would be helpful in the thoracic spine to avoid approaching both pleural cavity and in the lumbar spine to make easier and less dangerous the final maneuvers of specimen removal. The steps are the same as in type 3b, but after step III, once fully released the dural sac, an appropriate back to front oblique osteotomy is performed through healthy bone. It will be necessary to take care to cut in the right direction at appropriate distance from tumor margin and be sure of the protection of the anterior and antero-lateral structured, particularly the vascular structures. Navigation assistance is particularly helpful during this procedure. (D) Huge radio-induced osteogenic sarcoma in a 27 years old man. The previous tumor was a Hemangioendothelioma submitted to intralesional excision and radiation 5 years before. *En bloc* resection by type 3c technique. Transverse cut of the specimen including the plate and screws previously implanted. The margins were wide all over the tumor except in proximity of the pedicle (sector 9) where the tumor was found in the epidural space (intralesional margin). WBB, Weinstein-Boriani-Biagini.

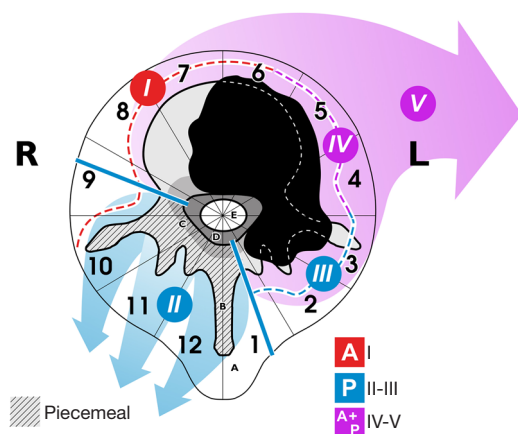


Figure 6 Type 6 WBB-based *en bloc* resection. A double anterior approach is useful for a safe release the aorta/cava bifurcation, in combination with a posterior approach to perform *en bloc* resection of a tumor located at L5. Proposed technique: first step in lateral position: antero-lateral approach on the side opposite to the tumor, release the homolateral aorta/cava bifurcation and partial discectomies or osteotomies to define upper and lower margin (I). Second step in prone position, same as type 5. The third step in lateral position. Re-opening of the posterior approach and contemporary retroperitoneal approach, release the homolateral aorta/cava bifurcation. Step IV: the psoas is transected to provide a margin over the tumor; step V: finalize discectomies or osteotomies to complete the *en bloc* resection and remove the specimen by the anterior approach. WBB, Weinstein-Boriani-Biagini.

in multilevel fusions, in order to avoid deformity, pain and instrumentation failures.

En bloc resection in the spine is therefore a very demanding surgical procedure, requiring oncological training and a team approach.

Indication and planning should follow some rules dictated by expert opinion and literature:

Diagnosis and staging must suggest that en bloc resection is the procedure of choice

Since 30 years the Enneking staging system has been adopted in many tumor centers and many reports and review confirm its validity. *En bloc* resection is “Enneking appropriate” for benign aggressive (stage 3) and for low grade malignant tumors (stage I). For high grade malignant tumor, *en bloc* resection is a valid option but must always be associated with chemotherapy or radiotherapy according to

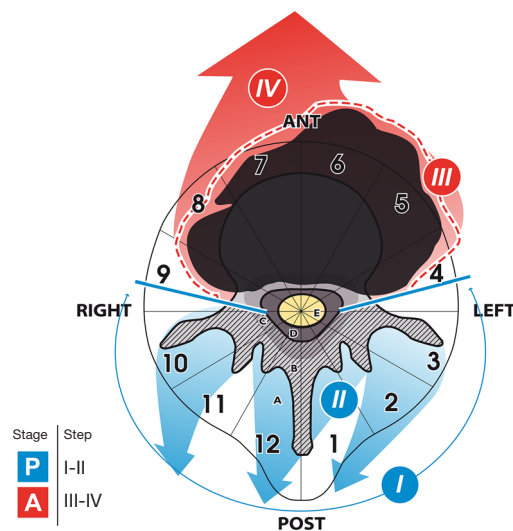


Figure 7 Type 7 WBB-based *en bloc* resection. It is indicated in thoracic and lumbar tumors which are growing anteriorly—even huge masses—in layer A without involvement of the canal (layer D) but with extension to layer C (in close proximity the posterior wall) and without involvement of sectors 4 and 9. This strategy allows to remove huge tumors without torsion around the spinal cord, but requires both pedicles free from tumor for an appropriate margin. It is mandatory to achieve by posterior a full release of posterior anatomical elements and spine-dura connection as in supine position no access will be possible. First steps in prone position: piecemeal excision of the posterior arch and both pedicles. Very careful full dura release. Discectomies or transversal grooves in vertebral bodies are performed to define the upper and lower margins. Second stage in supine position. Step III is release of the anatomical structures from the tumor mass or even their sacrifice to provide appropriate margin under visual control. Arterial by-pass can be performed in case of aorta involvement. The specimen is finally removed by combined maneuvers after complete upper and lower discectomies or osteotomies (IV). WBB, Weinstein-Boriani-Biagini.

the sensitivity of the specific tumor. Isolated spine metastases in patient in good general status, if not sensitive to radio and chemotherapy, can be considered for *en bloc* resection.

A tumor-free margin en bloc resection can be safely performed with acceptable functional loss if tumor extension and surgical anatomy fulfill the criteria of feasibility

Seven groups of strategy to plan *en bloc* resection have been proposed to define the criteria of feasibility of this

procedure according to tumor extension

Planning of the surgical procedure must consider the two previous points

The surgical approach or combination and timing of approaches must be decided combining the required oncological margins and the criteria of feasibility by tumor extension and by spine region.

If the margin is represented by relevant anatomical structures (dura, nerve roots, aorta, cava) a careful decision-making process will consider the improving of prognosis versus the functional loss. In this process the patient willing will be obviously relevant

Some details of surgical technique can reduce the complication rate and the morbidity

The morbidity profile of *en bloc* resections in the spine is high, due to the combination of the risks of anterior posterior spine surgery. Tumor surgery has also specific morbidity related to the need of dissecting through muscle and not through anatomical planes; further, *en bloc* resection require sacrificing not only the affected bone, but also almost all connecting elements creating a full instability.

Epidural bleeding should never be underestimated. Hemostasis is essential; poorly controlled epidural bleeding increases the risk of cardiovascular failure, post-operative hematoma, delayed wound healing, infection.

When the planning includes intralesional surgery or the risk of penetrating the tumor during resection is significant, selective arterial embolization is mandatory; however, when the surgeon anticipates a good probability of successful *en bloc* resection with oncological margins, tumor ischemia following embolization may induce peritumoral hyper-vascularization with increased risk of bleeding.

The final step of specimen removal must be planned to avoid tractions, torsions, shortening of the cord (19), particularly in multilevel resections (13). The effects on the cord vascularity during the tumor mass removal can be critical: it is one of the final step, after several hours of a bleeding surgery and the arterial pressure level should be kept still at a reasonable level to avoid the combination of stress and low blood flow.

The possibility that a single Adamkiewicz artery has the full responsibility of cord vascularity is controversial. Tomita and his group demonstrated on an animal model that the risk of cord ischemia is mostly related to the

number of contiguous radicular arteries sacrificed rather than to a single artery (20). It can be recommended to cut no more than three nerve roots bilaterally in the thoracic spine, and avoid acute shortening or distraction during the resection.

Electrophysiological monitoring has a role to guide permanent arteries occlusion.

As an obvious consequence to the requirement of tumor-free margins, anatomical compartments are frequently disrupted, as in case of *en bloc* resection of a thoracic spine tumor involving layer A: at the end, no barriers will exist through the peridural space and both pleural cavity if both parietal pleurae have been resected with the tumor for margin purpose. As a consequence, the post-op hematoma will develop around the dura and inside both pleural cavities. A cerebrospinal fluid (CSF) leakage following unrepaired or incompletely repaired dural tear will develop as a transpleural CSF fistula, seldom evolving towards self-repair due to the negative pressure existing in the pleural cavity. Surgical repair of the dural tear, dura patch, or even major procedures like omentum flaps have been proposed for such an awful complication.

Previous surgery and previous RT increase the risk of complications related to dissection. Infection is particularly threatening, due to the compromised immune status of many of these patients. Late aortic dissection is reported mostly in multi operated cases including aorta release and submitted to monoportal high dose conventional RT. Mortality rate can be relevant, till 2.2% (16).

Non-union is not rare among late complications due to the hostile environment to solid bony fusion. Vascularized graft has been proposed for safe fusion.

Excellent results however can be obtained by circumferential reconstruction achieved by connecting the cage to the posterior systems and by the use of carbon fiber composite systems, biologically active in promoting bone formation.

Acknowledgements

The author is indebted with Carlo Piovani, for his invaluable work: he translated into original images the concepts of individual approach to *en bloc* spine resection.

Footnote

Conflicts of Interest: The author has no conflicts of interest to declare.

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Cite this article as: Boriani S. *En bloc* resection in the spine: a procedure of surgical oncology. *J Spine Surg* 2018;4(3):668-676. doi: 10.21037/jss.2018.09.02