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Total en bloc spondylectomy for the L5 metastasis of a carcinoid tumor: illustrative case

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BACKGROUND Total en bloc spondylectomy (TES) was designed to achieve oncological complete tumor resection in a vertebral compartment. Because of the special anatomy of the lumbosacral junction, TES procedure at the L5 level is a challenge, and it has been explained in few reports in the literature. Performing TES in the lower lumbar region, as normal, is accomplished by using a combined approach.

OBSERVATIONS The authors presented the case of a 20-year-old man with an isolated spinal metastasis at the L5 level of carcinoid tumor of jejunum, limited to the vertebral body. Due to good long-term prognosis, after multidisciplinary evaluation the authors decided to treat the patient with TES through a combined posteroanterior approach, with posterior instrumentation and anterior reconstruction. Nine years after surgery, the patient was asymptomatic, with no sign of local recurrence.

LESSONS TES is a feasible technique to provide long-term survival in a select subgroup of patients, reducing the risk of local recurrence. The authors presented some anatomical and biomechanical factors that must be considered at the lumbosacral region. Despite the high rates of complication associated with TES, most patients benefit from local control provided by the technique.

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KEYWORDS lumbar vertebrae; spondylectomy; metastasis; neuroendocrine tumor

Metastatic carcinoma is the most common malignant bone tumor.[1] Tumors that tend to metastasize frequently to the spine are breast, lung, renal, prostate, thyroid, melanoma, myeloma, lymphoma, and colorectal cancer.[2] The incidence of bone involvement in neuroendocrine tumors has been estimated at 10%;[3] specifically, spine location is rare,[4–6] approximately less than 2%.[7] The curettage or piecemeal excision of vertebral tumors has been commonly practiced, but these approaches can result in incomplete resection of the lesion and high local recurrence rates, with poor results and prognosis for patients. Vertebrectomy and corpectomy for spine tumors resection involve intralesional removal of tumorous tissue and are not designed to remove all cancerous spine tissue in a gross total fashion.[8] On the other hand, total en bloc resections are designed to surgically remove a tumor in a single, intact piece, involved completely of healthy tissue,[9] without contaminating neighboring structures. In the last two decades, many authors[10–14] have described total corpectomy or spondylectomy in the spine as more aggressive techniques for decreasing local recurrence of vertebral tumors, with excellent clinical results. Total en bloc spondylectomy (TES) is an effective option for selected patients with no disseminated spinal tumors, and the technique provides improvement of functionality and increase in survival.[15–18] This procedure is considered one of the most demanding surgeries and requires a consolidated experience.[15] In this case report, we discuss a patient with a metastatic carcinoid tumor from the jejunum, with multiple abdomen metastases and a solitary vertebral metastasis in L5. A total en bloc L5 spondylectomy was performed using a combined posteroanterior double approach, in the context of a multidisciplinary treatment, to facilitate local control and longer disease-free survival.

Illustrative Case

History and Presentation

A 20-year-old man arrived at our hospital, presenting with an 8-year history of flushing and no additional symptoms, except tachycardia during the flushes. He reported an increase in the number and intensity of flushing episodes in the last 3 months as well as occasional mild low back pain. No abnormal findings presented on physical examination. Magnetic resonance imaging (MRI) and body

ABBREVIATIONS CT = computed tomography; MRI = magnetic resonance imaging; TES = total en bloc spondylectomy.
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computed tomography (CT) demonstrated multiple lesions in the abdomen: three in the liver, three in the mesentery, and one in the jejunum; the last one was suggestive of primary tumor. MRI and CT scans of the lumbosacral spine showed a single lesion involving the L5 vertebral body that did not extend to the posterior arch of the vertebra. ^{111}In-pentetreotide scintigraphy confirmed the nature and localization of the primary tumor and its metastases, and bone scintigraphy did not show any other bony metastases (Fig. 1). The patient received percutaneous CT-guided biopsy of the L5 vertebral body that demonstrated the mass to be consistent with a metastasis from a carcinoid tumor. The patient's Tokuhashi score (12 points) and Tomita score (1 points) indicated a prognosis of more than 6 months' survival.[15–17] Given the diagnosis and prognosis, TES was recommended after multidisciplinary evaluation.

Operative Procedure

The decision was made to resect the L5 vertebra via en bloc spondylectomy followed by spinal reconstruction to remove all tumoral tissue. We designed the operation to be performed in two stages over 2 separate operating days. Two days prior to the first stage, the patient received transarterial embolization of the tumor via L4 and L5 arteries as well as the middle sacral artery.

First Stage

The first surgical stage involved in en bloc resection of the posterior elements was followed by placement of instrumentation and fusion. The patient received general anesthesia and was positioned prone on the operating table. Continuous neuromonitoring was used throughout the procedure, and preoperative antibiotics were administered. A midline incision was made from the L3 to S3 level.

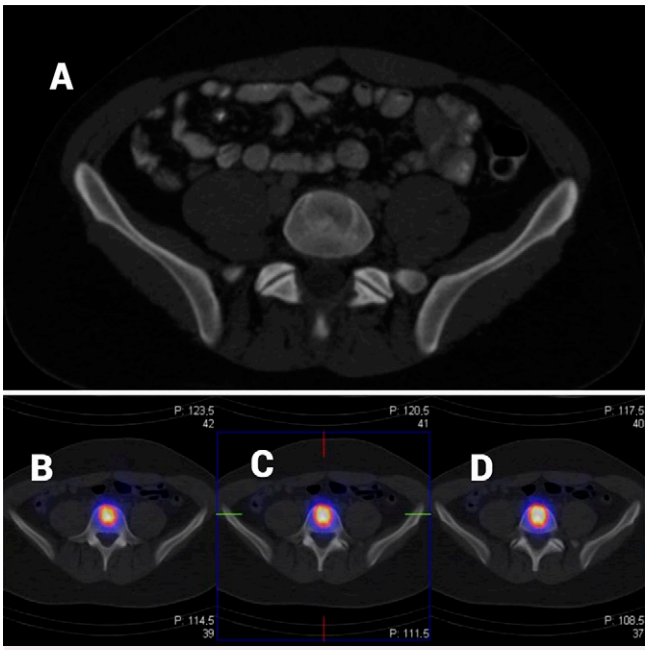

FIG. 1. A: Axial image of CT through the L5 vertebral body showing an isolated anterior-sided lesion in the vertebral body, with no effect on the posterior arch. **B–D:** Axial images of the ^{11}In-pentetreotide scintigraphy showing pathological caption of the tracing in L5 vertebra, compatible with tumoral effects with somatostatin receptors.

The spinous processes, lamina, facets, and transverse processes of L3 to L5 levels were carefully exposed in subperiosteal fashion, and the sacrum was exposed up to the sacral ala bilaterally. Pedicle screw instrumentation (Expedium, DePuy Synthes) was placed bilaterally from L3 to L4 and from S1 to iliac from the posterior superior iliac spine to the anterior inferior iliac spine. We carefully removed both sides of the L5 vertebral arch using a T-saw and exposing the thecal sac from L4 to S1. Once the posterior elements were removed, L4–L5 and L5–S1 discectomies were performed with division of the entire posterior longitudinal ligament between L4 and L5 and L5 and S1. Rods curved to fit the lumbar lordosis were connected to the L3, L4, and S1 pedicle screws and, via offsets, to the iliac screws. Demineralized bone matrix was placed after decortication of the facet joints and the transverse processes of L3 and L4, the sacral alae, and iliac crests. We washed the surgical site with saline, placed a small patch of Tachosil to secure hemostasis, inserted a drain, and closed the wound in multiple layers in the usual fashion. The pathological specimens of the posterior arch were revealed to be free of tumor in the anatomopathological analysis.

Second Stage

The second stage of the procedure was performed 8 days after the first stage. The patient was positioned supine under general anesthesia, and a midline combined supraumbilical and infraumbilical incision was made by a general surgeon, who removed the primary tumor from jejunum and performed a side-to-side anastomosis. To expose the anterior aspect of the lumbosacral junction, the inferior vena cava, the distal aorta, and the common iliac veins and arteries were dissected. Then we dissected around the L5 vertebral body and completed the L4–L5 and L5–S1 level radical discectomies, and the L5 vertebral body and tumor were removed en bloc (Fig. 2). For the anterior lumbosacral reconstruction, we inserted a titanium cage for vertebral corpectomy (Howmedica, Stryker) with 15° of lordosis, filled it with allograft bone and demineralized bone matrix, and fixed it in place (Fig. 3). Before closing the wound, a Gore-Tex mesh was placed to separate the cage from the surrounding tissue. A Jackson-Pratt drain was placed, and the wound was closed in the usual fashion.

Histological Analysis

The pathological specimens were determined to be jejunum carcinoid tumor and a metastatic lesion confined to L5 vertebral body. Margins of the pathological specimens were negative for tumor, as was the posterior arch. In accordance with current concepts of nomenclature about neuroendocrine tumors,[19] the final diagnosis and staging indicated a metastatic carcinoid tumor from the jejunum T3N1M1 (stage IV). The immunohistochemistry study found positivity for CD56, Ki-67 ($<$3%), chromogranin, and cytokeratin AE1/AE3 and negativity for CDX2 (Fig. 4).

Results

Postoperative Course

The patient was neurologically asymptomatic after surgery. No perioperative complications occurred, and the patient was discharged in stable condition after controlling blood loss that occurred intraoperatively. A postoperative CT scan revealed placement of posterior instrumentation from L3 to iliac and suitable placement of the L5 cage. Two weeks after surgery, the patient received hepatic transarterial chemoembolization for better control of the disease.

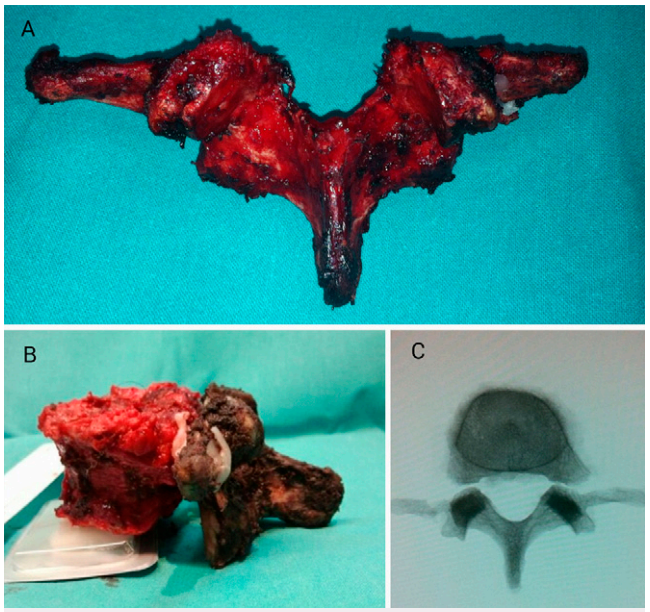

FIG. 2. A and B: Complete piece removed via spondylectomy, including the vertebral body and posterior arch (spinous process, bilateral lamina, superior and inferior articulating processes, bilateral pars, bilateral transverse processes, and bilateral pedicles) separately. The anatomopathological analysis showed a lesion of 2.5 × 2 cm, proximal to the anterior rim of the vertebral body, without effect of the surgical margins. The analysis was compatible with a neuroendocrine tumor metastasis. At the posterior arch, the analysis did not reveal histological signs of metastatic effect. **C:** Radiographic study of the complete piece removed via spondylectomy.

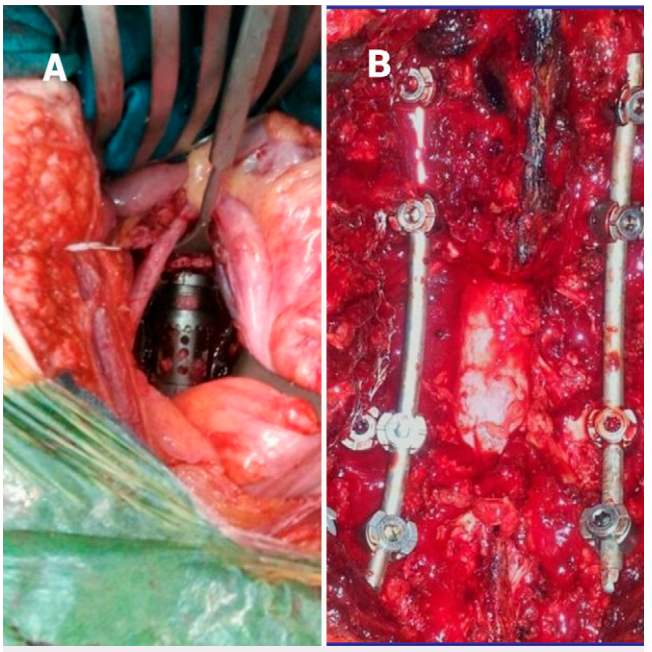

FIG. 3. A: Anterior reconstruction with a cage after resection of the vertebral body, with 15° of lordosis, with a Gore-Tex mesh to separate it from all the tissues around. **B:** Posterior instrumentation placed from L3 to the iliac after resection of the posterior arch. Rods curved to fit the lumbar lordosis were connected to the L3, L4, and S1 pedicle screws and, via offsets, to the iliac bone screws.

For 8 weeks, the patient wore a lumbosacral orthotic device for comfort and pain control.

Follow-Up

The patient is now 9 years out from surgery. Lumbosacral CT scanning on follow-up after 6 years showed no breakage of the instrumentation, no cage displacement, no radiolucent lines around the pedicle screws, and the maintenance of spinal alignment. There has been no evidence of bone metastases or tumor recurrence in the spine to date. The latest radiographs obtained approximately a year ago showed no alterations (Fig. 5). The patient has a good quality of life and a good long-term prognosis.

Discussion

Observations

TES was put in practice in the 1990s by Tomita.[14–17] Numerous authors[15,20,21] have increased their interest in this surgical treatment, and total excision and en bloc resections have provided improvement in the prognosis for patients.

It aims to achieve oncological complete tumor resection considering also the satellite microlesions in a vertebral compartment to avoid local recurrence.

Enneking et al.[22] suggested the conceptualization of "compartment and anatomic barriers." Applying this to the spine, Tomita et al.[15] concluded that one vertebra could be considered a single oncological compartment. We must take into account the anatomical and biomechanical considerations at the lumbosacral region that increase the level of difficulty of the technique compared to the thoracic spine.

Indications

According to the punctuation system by Tomita et al.,[16] vertebrectomy is indicated in patients with slow-growing tumors, without visceral metastases and solitary and isolated vertebral lesions, and with almost 2 years of life expectancy. Therefore, only a small number of patients are considered possible candidates. Patient selection must be undertaken according to the following two factors:

1. Local vertebra tumor: Tomita et al.[23,24] devised a surgical classification based on both the pattern of local vertebral tumor progression and the type of surgery used to excise it. Vertebrectomy is recommended in intracompartmental lesions (types 1, 2, or 3), particularly when the vertebra is cut at the healthy part of pedicle or lamina. For types 4, 5, and 6, a marginal margin may be possible only if the lesion is well encapsulated with a fibrous reactive membrane. Nevertheless, the indication must consider clinical and radiological characteristics of each case.[18]
2. Preoperative evaluation: Tokuhashi et al.[17] proposed a prognostic scoring system for preoperative evaluation of patients with metastatic spine tumors. Tomita et al.[16] designed a scoring system with three prognostic factors, which are regarded as the most influential factors for life expectancy: the pathological/clinical grade of malignancy, the presence of visceral metastases, and bone metastases. According to this score, TES would be indicated in patients with 2 to 3 points and a life expectancy longer than 2 years.

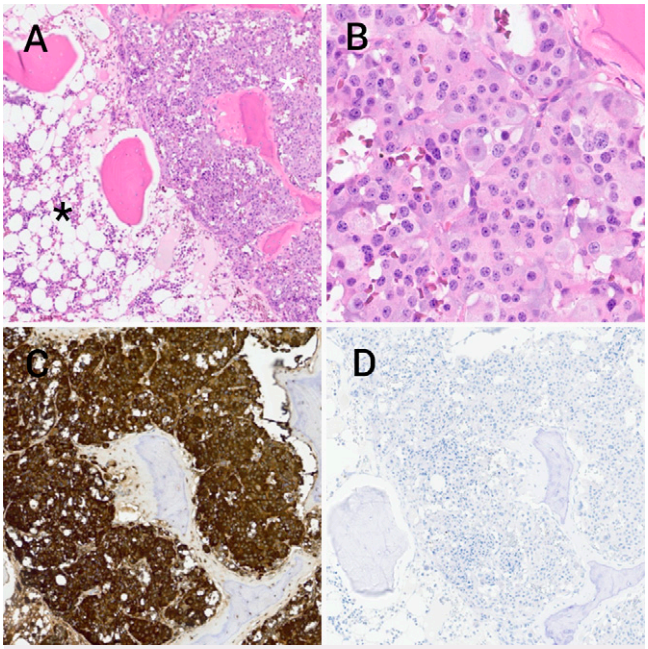

FIG. 4. A: Bone metastasis of a neuroendocrine tumor. Hematoxylin and eosin (H&E), original magnification ×8.5. Black asterisk indicates free tumor bone marrow. White asterisk indicates tumoral tissue. It can be appreciated in a solid-infiltrative pattern, without necrosis. **B:** Neuroendocrine tumor. H&E, original magnification ×40. Note the uniform cells, extensive cytoplasm, and central round nucleus, in which salt-and-pepper chromatin appears. Observe the low cellular pleomorphism, without mitosis or prominent nucleolus. **C:** Immunohistochemistry study with chromogranin (original magnification ×12), a characteristic sign of neuroendocrine tumors, was intensely positive. **D:** Immunohistochemistry study with Ki-67 (original magnification ×12). The proliferation index was <3%, so in addition to the low mitotic index observed, we could classify the lesion as a low-grade tumor (grade 1).

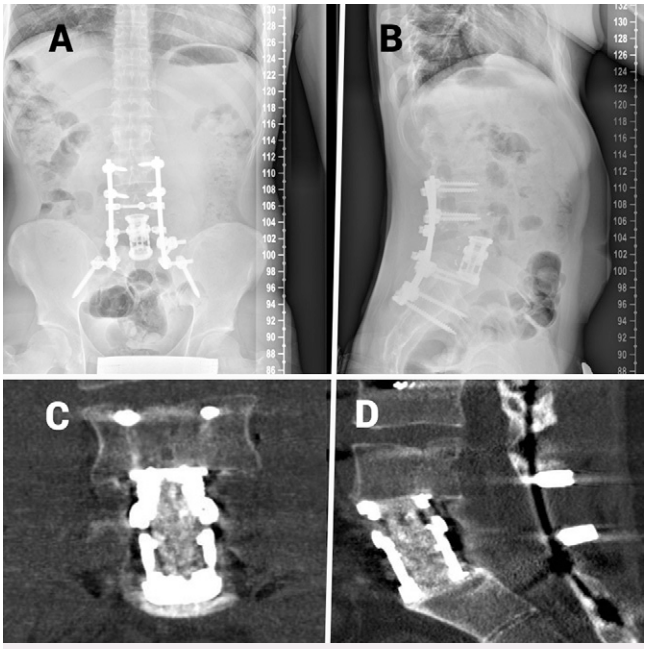

FIG. 5. Anteroposterior (**A**) and lateral (**B**) views of a telemetry 9 years after surgery, with no signs of implant release. Coronal (**C**) and sagittal (**D**) reconstructed CT scans obtained 6 years after surgery, showing placement of the interbody cage, pedicle screws, and rods with no signs of instability and good alignment in the sagittal plane.

Taking in account these considerations, our case represents a patient with an isolated vertebral lesion at L5 level, type 1 according to Tomita's classification. It is true that our patient presented with visceral metastases at the time of diagnosis (three in the liver, three in the mesentery). However, due to the good prognosis of these metastases and the possibility of developing a curative treatment, we decided to implement the TES procedure. The patient had 12 points according to the Tokuhashi prognosis score system, so he was a good candidate for the procedure. When there is distant metastasis on the bone, the 5-year survival decreases from 67% to 4%–35%.[25] No studies have yet compared other options of treatment with TES, so it is important to have more evidence to determine appropriate option for each patient. The rate of local and distant recurrence of the spine metastasis in carcinoid tumors is high,[25,26] so we opted for spondylectomy, a more aggressive treatment, to provide a good long-term prognosis, taking into account that in our patient we had the option of completely removing the primary lesson in the jejunum with adequate margins.

Preoperative Considerations

It is important to reduce excessive bleeding, which occurs sometimes in patients with hypervascular tumors, like the case we present (carcinoid tumor of jejunum). A few studies[15] have reported that in these patients, preoperative embolization of the feeding artery at the affected vertebra is shown to reduce intraoperative bleeding without compromising spinal cord function. Maintaining systolic blood pressure in low values (80-100 mm Hg) and the use of hemostatic techniques to reduce epidural bleeding have been shown to reduce excessive bleeding. In our hospital, we use fibrin patches such as Tachosil, and in the case we report, transarterial embolization of the tumor was performed via L4 and L5 arteries as well as the middle sacral artery before performing the TES procedure.

Choice of Approach

Surgical approaches to the L5 vertebra entail difficulty due to anatomical characteristics of the lumbosacral junction. For spinal tumors at the L5 level, there is consensus in the literature[15,27,28] about using a combined posteroanterior procedure. This strategy typically is accomplished by a posterior resection of the involved vertebra and instrumentation followed by anterior corpectomy and positioning a vertebral prosthesis (either in a single or staged procedure). The reason for this is the complex anatomy represented by the iliac wing and lumbosacral plexus nerves and the size of the L5 vertebral body, which may cause damage of lumbar nerve roots when vertebra are removed.[24,29] D'Aquino et al.[28] studied in a systematic revision the different surgical approaches to L5 corpectomy and reconstruction. Three studies evaluated outcomes after a combined posteroanterior performance; one study followed anterior-only corpectomy and the other two followed a posterior-only approach. More than half of the surgeons involved a combined surgical approach. They concluded that the combined approach is frequently linked with longer operative time, high blood loss, and morbidity. Nevertheless, the treatment was

successful, regardless of surgical approach. In our case, we describe a two-stage operative procedure. We undertook TES and lumbosacral reconstruction via a combined posteroanterior approach in two different days, using the second surgery for both removing the primary tumor (by general surgeon through a laparotomy) and performing the anterior approach to the spine. The use of a posterior-only approach is technically challenging for L5 tumors, although Li et al.[30] reported a case (62-year-old woman with breast cancer and a single lesion in the L5 vertebra) without severe complications. After that, Yang et al.[31] also reported satisfactory results and good tumor control for a few L5 tumors using a posterior-only approach.

Consequently, the surgical approach varies depending on several factors, such as the primary tumor and surgical objectives and the level of surgeon experience.[28]

Technique Considerations at the L5 Level

Regarding the safest interval to deal with this procedure, Sangsin et al.[29] used the interval space between the left common iliac vein and artery during the anterior stage, and Gallia et al.[32] used the space between the bifurcation of the common iliac vessels. Because of the biomechanics of the lumbosacral junction (characterized by a sloping transition from a dynamic lower lumbar region to a relatively motionless sacrum and pelvis), there are stabilization problems (the L5 vertebra is affected by sliding and compressive forces).[27,28,33] Therefore, it is important to achieve enough stabilization, supporting the necessity to provide 360° of spine stabilization. In this region, the anterior or posterior approaches may be insufficient to ensure this stabilization, so the use of a double posteroanterior approach is recommended.[27] In our case, to accomplish a good anterior and posterior stabilization, we performed a posterior pedicle bilateral screw instrumentation from L3 to L4 and from S1 to iliac as well as an anterior reconstruction with a cage filled with allograft bone and demineralized bone matrix fixed in place. Vazan et al.[33] reported in their study three cases of anterior cage dislocation with a combined anteroposterior surgery. They demonstrated a significant difference in mean postoperative local lordotic angle, concluding that a high postoperative local lordotic angle $>50°$ was a potential risk factor for construct failure. The mean local lordotic angle in the nonfailed group was 32.1° (range, 21.1–46.2; $p = 0.0026$). In our case, we fixed the cage with 15° of lordosis without signs of dislocation in the long-term postoperative period.

Complications

En bloc resection is a procedure with high morbidity, which must be considered in the decision-making process. It is important to take into account some considerations that may influence the success of the procedure: preoperative treatments such as radiotherapy or chemotherapy, which may increase the risk of local complications; the immunosuppressed status of the patient; hypervascular tumors, which may cause excessive blood loss; tumor location (high demanding procedure at L5 level with the possibility of need for double posteroanterior approach); and the number of resected segments. In a retrospective study, Boriani et al.[9] investigated 220 en bloc resections with occurrence of complications. In their study, the main risk factors identified were the condition of important structures after previous surgery or radiation, lack of total control of blooding loss, double combined approach, and too short posterior fixation and lack of anterior support. Nevertheless, it is necessary to take into account that a less aggressive surgery may cause inappropriate margins, increasing the risk of local recurrence and worse patient prognosis. However, the literature shows that the complication risk is balanced by better local control of the tumor and good long-term prognosis.[9]

Lessons

Good results and survival rates using TES have been described in numerous studies and reports,[12,15,20,23,24,34–38] and from Tomita's studies,[15,16,23,24,34] this technique increased interest among spine surgeons regarding management of spine metastases, not only by treating local lesions but also by improving patient prognosis and avoiding local recurrence of tumor. Vertebrectomy provides an increase in life expectancy compared with curettage or piecemeal excision. The technique also improves quality of life, and most patients preserve or improve their neurological status.[18] Good local control justifies performing these procedures in aggressive benign and low-grade malignant bone tumors.[9]

Because of the special anatomy of the lumbosacral junction, performing total spondylectomy at the L5 level is a challenge, and it has been described in a few reports in the literature.[36] In our case, after careful multidisciplinary discussion and evaluation, TES was indicated as a feasible technique to reduce the risk of local recurrence. Although the liver metastases of the patient are still not completely controlled, there is no evidence of bone metastases or tumor recurrence in the spine at the present time, so we can say that the results are excellent after the spondylectomy. In conclusion, TES with spinal reconstruction is a safe and effective treatment for solitary metastasis and can provide long-term survival in particular circumstances.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Jódar, Fuentes Caparrós. Acquisition of data: Jódar, Fuentes Caparrós, Osuna Soto. Analysis and interpretation of data: Fuentes Caparrós, Osuna Soto. Drafting the article: Jódar, Fuentes Caparrós. Critically revising the article: Fuentes Caparrós. Reviewed submitted version of manuscript: Jódar, Fuentes Caparrós, Osuna Soto. Approved the final version of the manuscript on behalf of all authors: Jódar. Statistical analysis: Marín. Administrative/technical/material support: Jódar, Fuentes Caparrós, Osuna Soto. Study supervision: Fuentes Caparrós, Osuna Soto.

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