

Total *en bloc* spondylectomy of T11 and spine shortening performed on a 17-month-old patient: art of the possible

Laurent Coubeau¹ · Cecile Boulanger¹ · Frederic Lecouvet¹ · Mo Saffarini²  · Xavier Banse¹

Abstract

Study design Case report.

Purpose The authors used spine shortening as an alternative strategy to intercalary graft fixation to restore permanent spine stability for a 17-month-old infant who received total *en bloc* spondylectomy (TES) of T11 to treat an embryonic rhabdo-myosarcoma. TES involves complete removal of vertebra, compensated by spine reconstruction using intercalary allografts and permanent posterior instrumentation, which is not possible for skeletally immature patients with high growth potential and non-ossified vertebrae.

Methods Surgery was performed over two consecutive days. During the first day, the tumor was released from its dorsal attachments through the posterior approach. During the second day, the tumor was dissected and excised through the ante-rior approach, leaving a gap between T10 and T12. The two vertebrae were then drawn toward each other until the gap was bridged. The dural sac slipped into the canal under T10 and T12 with no observable kinking.

Results Fifteen weeks after surgery, thoraco-abdominal CT confirmed fusion of the T10 and T12 vertebral bodies. Three years later, the patient lives a normal life with no major neurological deficits or recurrence of sarcoma.

Conclusions This case report is the first to demonstrate the feasibility of TES with spine shortening of an entire thoracic segment without spine kinking or damage in an infant. This unprecedented surgical technique allowed complete removal of an embryonic rhabdomyosarcoma, while granting rapid stability and growth potential.

Level of evidence IV.

Keywords Spondylectomy · Spine shortening · Pediatric surgery · Vertebrectomy · Oncology · Rhabdomyosarcoma

Introduction

The proximity of spinal tumors to the dural sac renders their excision challenging. Prognosis often depends on the ability to remove the tumor entirely without intralesional resections. Total *en bloc* spondylectomy (TES) is the preferred option to treat tumors of the thoraco-lumbar spine [1] and involves complete removal of one or more vertebrae, compensated by reconstruction using intercalary allografts and instrumentation [2]. Surgical reconstruction is generally performed with allografts or cages in adult cases. This relies on relatively

long and stable instrumentation until the gap is filled by living bone and fusion is complete. In cases with high growth potential and non-ossified vertebrae, spine reconstruction using permanent instrumentation was found to be ineffective or impossible [3], as it does not provide sufficient immediate stability and may require multiple re-operations. Spine shortening was therefore considered, to directly increase stability (eliminating the intercalary component) and to sharply accelerate fusion, because it creates direct contact between the vertebral bodies. We report the unique case of an infant who had TES for rhabdomyosarcoma using an unprecedented salvage strategy, shortening the spine by an entire segment.

✉ Mo Saffarini
journals@resurg.eu

¹ Cliniques Universitaires Saint-Luc, Institut de Recherche Expérimentale et Clinique (IREC), Université catholique de Louvain (UCLouvain), Bruxelles, Belgium

² ReSurg S.A., 22 Rue St Jean, 1260 Nyon, Switzerland

Case report

A 14-month-old infant was admitted to our pediatric oncology department due to an extensive mass around his eleventh thoracic vertebra (T11), with histological diagnosis of embryonal rhabdomyosarcoma without metastasis (T3N0M0) (Fig. 1). The patient responded to neoadjuvant chemotherapy (RMS2005 protocol) which decreased lesion size by more than 50% within 3 months. However, preoperative thoracic CT scan confirmed that the tumor had invaded the T11 vertebral body, rendering spondylectomy inevitable (Fig. 2).

Surgical removal of the tumor by TES was planned, but due to the infant's growth potential and non-ossified vertebrae, instrumentation and allograft were not an option. The authors therefore considered spine shortening to restore stability rapidly, but to their knowledge, spine shortening by an entire segment was never performed around the spinal cord. After evaluation of risks and benefits, and considering the softness of non-ossified vertebrae, shortening the trunk by the entire T11 segment was deemed the best treatment option for this patient.

Surgery was performed under general anesthesia over two consecutive days. On the first day, the tumor was dissected from posterior tissues, including the entire T11, carefully separated from the epidural fat under a microscope. The spine was then instrumented, using four 12-mm-long pedicle screws with a core diameter of 2.4 mm and 3.5 mm rods, on T10 and T12, and additional four small hooks on T9 and L1 (Synapse[®] instrumentation, DePuy Synthes, Raynham, MA). One screw (T10 right) was replaced by a hook due to loss of screw stability after transverse process resection (Fig. 3). After closure of the incision, a wake-up test was performed to verify lower limb motor function. The patient was then transferred to intensive care and kept anesthetized overnight. On the second day, the tumor was dissected from anterior tissue (including aorta, vena cava and liver) and

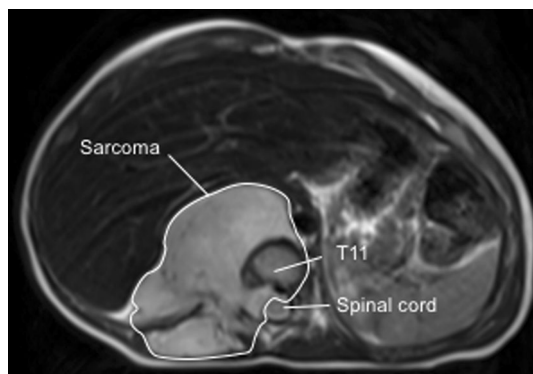


Fig. 1 Preoperative MRI section showing the sarcoma and T11

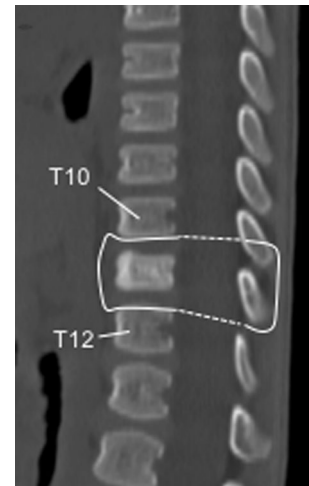


Fig. 2 Preoperative sagittal CT reconstruction revealing the invasion of T11 by the sarcoma

excised en bloc through the anterior approach, leaving a 1-cm anterior gap between T10 and T12. The posterior incision was reopened to expose the instrumentation. The two vertebrae were then drawn together along longitudinal titanium rods, until the gap was bridged. The dural sac slipped into the canal under T10 and T12 with no visible kinking, providing tight bony contact.

Analysis of the tumor specimen confirmed that resection was achieved without effraction. Five cycles of adjuvant chemotherapy were given over 4 months. At 4 postoperative weeks, the patient walked for the first time. At 15 postoperative weeks, there was no bulging of instrumentation at the skin, and thoraco-abdominal CT confirmed fusion of the T10 and T12

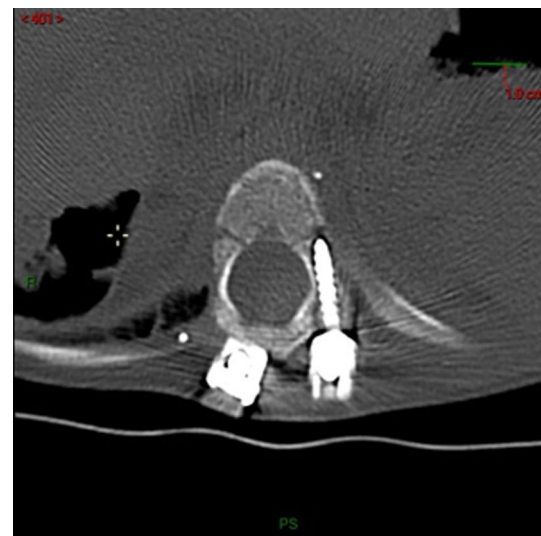


Fig. 3 Postoperative axial CT reconstruction showing the very small T10 left pedicular screw at 15 weeks. Note that the screw tip does not perforate the growth plate

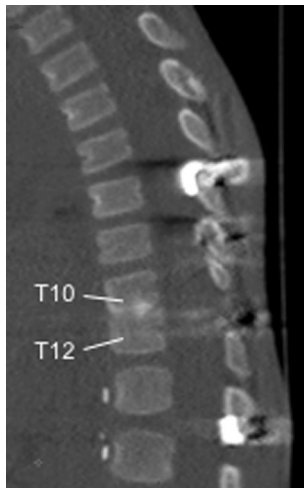


Fig. 4 Postoperative sagittal CT reconstruction demonstrating fusion of the T10 and T12 bodies at 15 weeks, just before removal of instrumentation

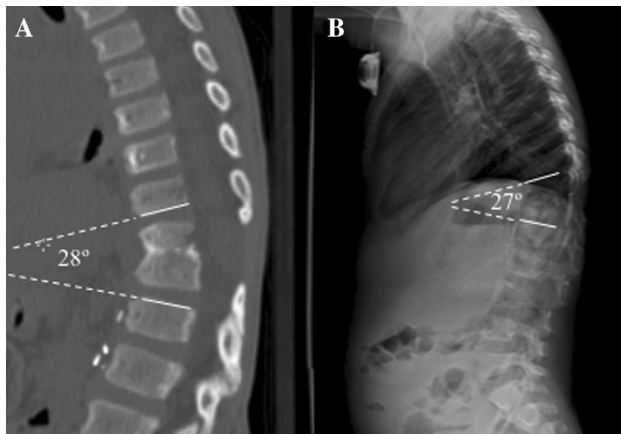


Fig. 5 Postoperative **a** sagittal CT reconstruction at 24 months confirming mature fusion between T10 and T12 with local kyphosis of 28° and **b** full spine standing radiograph at 42 months showing local kyphosis of 27° but no major deformity

vertebral bodies (Fig. 4), allowing removal of the instrumentation (Fig. 3). At 24 postoperative months, thoraco-abdominal CT showed mature fusion between T10 and T12, with a mild local kyphosis (28°). At 42 postoperative months, the patient has the same mild local kyphosis (27°) without major deformity in standing position and lives a normal life with no neural deficits or recurrence of sarcoma (Fig. 5).

Discussion

Standard TES with long instrumentation and reconstruction with intercalary allograft or titanium cage is problematic for infants with non-ossified vertebrae, as these

instrumented constructs cannot provide immediate stability and as the patients may require multiple re-operations due to their high growth potential [3]. In this report, we present the case of a 14-month-old infant operated with TES for an embryonic rhabdomyosarcoma, who had spine shortening by the entire T11 segment, as spine reconstruction was not possible. This unprecedented strategy restored immediate stability after TES without the need for posterior instrumentation, although long-term success is yet to be confirmed and the patient will require monitoring until adulthood. Neuromonitoring was not possible at our institution for this patient due to his small size which, over the span of 2 days, would be unreliable. Neuromonitoring would have otherwise been beneficial and should be considered for all future cases. Three-year follow-up is promising, since the mild observed kyphosis (27°) is carefully monitored and did not show any evolution over the last 18 months (Fig. 5). The kyphosis is mainly due to the absence of posterior bony elements and the complete excision of the paravertebral muscles and ribs on the left side of the trunk. Rapid hardware removal allowed an efficient MRI follow-up to inspect for local recurrence.

Spine shortening is sometimes performed to correct spine deformities or treat tethered cord syndrome [4, 5], but in such instances the spine is shortened by considerably less than an entire segment and fixed using permanent posterior instrumentation [2, 5]. Successful spine shortening by an entire segment has only been reported once, on a 13-year-old child, who had piecemeal resection of L6, followed by spondylodesis of L5 and S1 [6]. Subsequent reports highlighted the risks of spine damage associated with this method [7–9], and to our knowledge, spine shortening by an entire segment has never been performed at levels that surround the spinal cord.

The decision to shorten the spine was based on the patient's tissue softness and growth potential. There was no visible kinking of the dural sac during reduction, and subsequent neurological examination revealed no deficit. This patient is likely the youngest ever to receive TES and the first living with a spine shortened by an entire vertebral segment, at the spinal cord level.

Compliance with ethical standards

Conflict of interest LC, CB, AP, FL, MS and XB declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

References

1. Shimizu T, Murakami H, Demura S, Kato S, Yoshioka K, Yokogawa N, Kawahara N, Tomita K, Tsuchiya H (2018) Total en bloc spondylectomy for primary tumors of the lumbar spine. *Medicine* 97(37):e12366. <https://doi.org/10.1097/md.00000000000012366>
2. Tomita K, Kawahara N, Murakami H, Demura S (2006) Total en bloc spondylectomy for spinal tumors: improvement of the technique and its associated basic background. *J Orthop Sci* 11(1):3–12. <https://doi.org/10.1007/s00776-005-0964-y>
3. van Dijk M, Smit TH, Burger EH, Wuisman PI (2002) Bone histomorphometric evaluation of a clinically fused titanium tumour cage in a child. *Eur Spine J* 11(5):507–511. <https://doi.org/10.1007/s00586-002-0429-3>
4. Lin W, Xu H, Duan G, Xie J, Chen Y, Jiao B, Lan H (2018) Spine-shortening osteotomy for patients with tethered cord syndrome: a systematic review and meta-analysis. *Neurol Res* 40(5):340–363. <https://doi.org/10.1080/01616412.2018.1446268>
5. Yoshioka K, Murakami H, Demura S, Kato S, Kawahara N, Tomita K, Tsuchiya H (2013) Clinical outcome of spinal reconstruction after total en bloc spondylectomy at 3 or more levels. *Spine* 38(24):E1511–E1516. <https://doi.org/10.1097/BRS.0b013e3182a6427a>
6. Shimizu K, Ido K, Fujio K, Tanaka K, Nakamura T (1996) Total spondylectomy and spinal shortening for giant-cell tumour of spine. *Lancet* 348(9023):342
7. Kawahara N, Tomita K, Kobayashi T, Abdel-Wanis ME, Murakami H, Akamaru T (2005) Influence of acute shortening on the spinal cord: an experimental study. *Spine* 30(6):613–620
8. Modi HN, Suh SW, Hong JY, Yang JH (2011) The effects of spinal cord injury induced by shortening on motor evoked potentials and spinal cord blood flow: an experimental study in Swine. *J Bone Joint Surg Am* 93(19):1781–1789. <https://doi.org/10.2106/jbjs.I.01794>
9. Pedrals JG (1996) Spinal shortening in scoliosis surgery, a case with transitory paraplegia. *Spine* 21(21):2515–2519