Case Report

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Total En Bloc Spondylectomy of T11 Osteosarcoma with Dural Resection: Case Report and Literature Review

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Abstract

Wide margin en bloc resection is becoming the trend for the treatment of primary spinal tumors. However, only few cases in the literature describeen bloc tumor resection including dural resection. We here describe a case of wide en bloc resection of a spinal osteosarcoma using spondylectomyanddural flap resection, followed by duraplasty repair. This was followed by adjuvant chemotherapy. At forty-two-monthsof follow-up, the patient was stable neurologically and free of disease radiologically. The clinical presentation, operative technique and related literature are reviewed.

Keywords: En Bloc; Spondylectomy; Dural Resection; Duraplasty.

Introduction

Primary tumors of the spine are less common than metastatic lesions. Theirsurgical management creates challenges due to the anatomical complexity of the spine and the surrounding vital structures. Thiscomplexity makes it difficult to carry out en bloc resections with wide margins. For this reason, piecemeal intralesionaltumor excision has become a historical standard[1,2].With the development of imaging studies, and of spine surgical and instrumentation techniques,more invasive resection evolved to achieve adequate margins. Recently, wide margin resection is becoming increasingly popular, offering patients an outcome comparable to primary bone tumors of the extremities. Paraspinal muscles, ribs, biopsy tract, nerve roots, and less commonly some surrounding organs like kidney or lung lobe, dura, cauda equina and finally spinal cord have all been reported to be included in the en bloc resection of these tumors [1–7,8].

Including the dura in the en bloc resection with the intent of preserving spinal cord function has rarely been reported in the literature [4–7,9,10]. In this article we report a case of dural resection with preservation of neurologic function in primary osteosarcoma of the lumbar spine with a comparative review of similar reported cases.

Case History

A 30-year-old lady who was previously very active in outdoor sports began havingpain in the middle of the back with thoracic radicular radiation. There was no history of trauma apart froma remote incidence of falling on her back. She had a known remote 30% wedge compression fracture at L2 without clinical sequelae.Neurological examination was normal. Eventually, in a community hospital, she was diagnosed with a T11 right pedicle tumor (Figure 1). An open biopsy was carried out. After the diagnosis of osteosarcoma was made, a pedicle excision and partial vertebrectomy was carried out in the referring

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Table 1: Overview of reported cases of en bloc spondylectomy including the dura. (mo= month, yr= year, NED= No evidence of disease, AWD= Alive with disease, DOD= died of disease, BP= Back pain)

Author	Age/ sex	Clinical presentation	Level	Dura resection/ replacement	Pathology	Severed neural tissues	Follow up
Krepler[6]	27y,M	paraplegic	T4 to T7	Dorsal part at T5-T7. Reconstructed by lyophilized dura	Osteosarcoma. No microscopic details of dural involvement	_____	NED, 116 mo
Biagini[10]	61y,M	BP, neurogenic claudication and constipation	T12-L2	Dural "robe" technique (dual circumferential patch) using lypholized pericardium	Chordoma. No microscopic details of dural involvement.	T11-L2 roots	AWD, 36mo (had lung nodule before surgery, which was stable. No local disease)
Fisher[4]	36y,?	?	C5-T1	Extensive dural resection with patch repair.	Malignant primary nerve sheath tumor. No microscopic datails.	Right C7,C8,T1	DOD, 22 mo
Keynan[5]	20y,M	paraparesis + numbness from L2 distally. Decreased rectal tone and perianal sensation.	L2	From T12 to L4 circumferentially+ completely transecting the conus and cauda equina at the top end and at the bottom end	Osteosarcoma. Tumor adherent to dura. But didn't violate subarachnoid or pial spaces.	Conus, cauda at L1-L3 and corresponding roots	NED, 4y
Muraka -mi[7] Case 1:	21y,M	paraplegic	T1 to T5	From C7 to T6 circumferentially	Telangiectatic osteosarcoma. Tumor adherent to dura and destroyed its outer layer in part. No pseudomembra ne or dural invasion.	Spinal cord at T1-T5 and corresponding roots	DOD, 6mo
Case 2:	57y,M	paraplegic	T11 to L1	T11 to L1 circumferentially	Telangiectatic osteosarcoma. Tumor adherent to dura but no destruction , invasion or even pseudomembra ne	Spinal cord at T11-L1 and corresponding roots	DOD, 4mo
Aryan[9]	60y,M	Back pain	L3-L4	Dorsal dura removed, patch repair.	Renal cell carcinoma. Dura grossly infiltrated. No microscopic details.	_____	NED,6mo
Chanpla -korn[18]	14y,M	BP, paraplegic, no sacral sparing	T11-L4	T11-L4 circumferentially with resecting the conus meullaris and cauda equina/ running suture for dural stump	Recurrent epithelioid sarcoma, free margins. Gross epidural invasion, no microscopic details	Conus and cauda at T11-L4	AWD (suspicious new lung consolidation), 26mo
current	30y, F	BP, radiculopath y	T11	Right lateral durotomy, patch repair.	High grade osteogenic sarcoma. Dura grossly intact, no invasion.	Right T11	NED, 17mo

hospital, through the same tract. The patient was subsequently referred to the musculoskeletal oncologist at our center for further treatment. Histopathology at that time was suggestive of high-grade osteogenic sarcoma. A CT scan showed a T11 vertebral body lesion and evidence of right pediculectomy with right hemilaminectomy and partial right corpectomy (Figure 2). An MRI showed the biopsy tract extendingfrom the lower border of T9 to the upper border of T12. There was a suspicion of dural involvement on the right side (Figure 3 and Figure 4). The canal was otherwise free of tumor extension. Extensive staging work-up was negative for other lesions. Previous interventions carried out at an outside hospital were suspected to be all intralesional.

Treatment Plan

We believed that all previous interventions were intralesional. Threfore, neoadjuvant chemotherapy followed by resection of the previous surgical tract along with an en bloc T11 vertebrectomy was planned. The chemotherapy included 3 cycles of Adriamycin and Cisplatinum. Dural excision was planned if the dura was found at surgery to be adherent at the surgical site.

Operative Procedure

Bilateral transpedicular screws were inserted at the levels of T8, T9, L1 and L2. At T10 only a left sided pedicular screw was inserted, as the right pedicle was partially resected during the surgery. The rods were contoured in accordance to her anatomical spinal curvature and then removed. One-stage posterior total en bloc T11 spondylectomy was then done, including the resection of the primary biopsy tract (Figure 5). Intra-operatively, the dural sac was obviously adherent to the right side at the level of T10 and T11 pedicles. This was thought to be caused either

Fig. 1: Sagittal (left) and axial (right) CT showing right T11 pedicle prior to all surgeries with the associated osteoblastic lesion seen just inferior and medial to the pedicle

Fig. 2: Axial (left) and sagittal (right) CT showing right T11 pediculectomy, laminecomy and partial corpectomy in the primary biopsy procedure. Residual unresected tumor appears as a lytic lesion in the body of T11 (right)

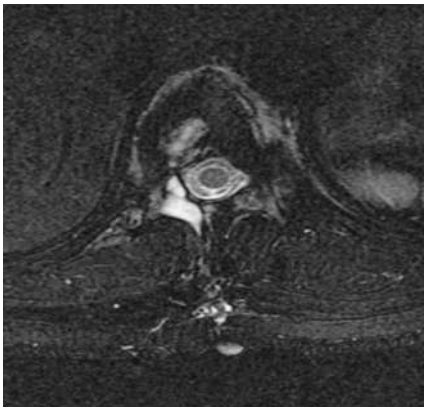

Fig. 3: Enhanced Axial T1 MRI showing post-operative changes following biopsy and then intralesional pedicle excision

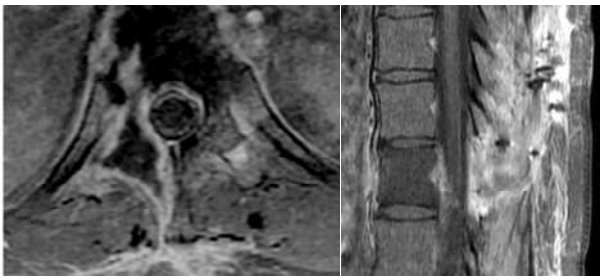

Fig. 4: Sagittal T1 (left) enhanced images. Axial T1 enhanced (right) images following the first 2 surgeries (biopsy and then intralesional pedicle excision)

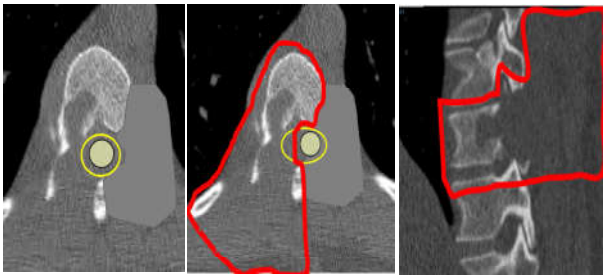

Fig. 5: the surgical procedure illustrated on top of the preoperative CT. Step1- resection of a lateral opening for the cord (left) from the left side, which was unviolated from the previous operations. Step 2- Axial and sagittal views (middle and right) for the dissectionalong the en bloc margins including the adjacent rib, the previous surgical corridor and the adherent dura onthe right side

by the tumor or secondary to the previous surgery. Giving this finding, the decision was taken to proceed with the planned dural resection including,intradural ligation of the right T11 nerve root followed by duraplastyusingxenograft patch. A cage was applied at the level of T11 and the rods were reinserted (Figure 6). Neither the tumor nor the prior surgical site was visualized during the entire operation. The total anesthetic time was 10 hours and the estimated blood loss was 7.5 liters.

Histopathology

There was no microscopic tumor contamination at the surgical margins and uninvolved overlying dura and bony cortical surfaces. The specimen was otherwise benign with absence of viable tumor cell. That was interpreted as an evidence of complete response to chemotherapy (Figures 7 & 8).

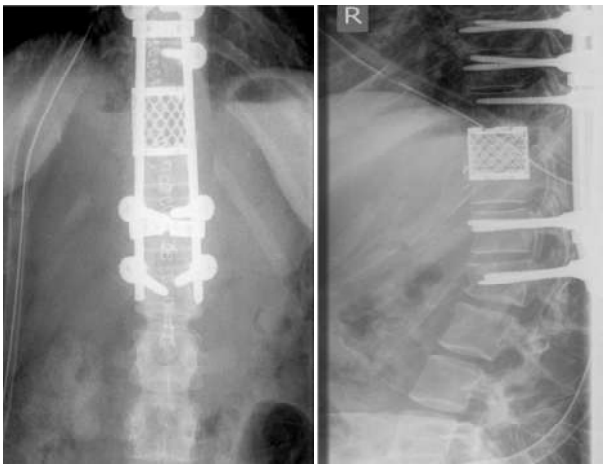

Fig. 6: AP (left) and lateral (right) X-ray showing post-operative anterior and posterior instrumentation

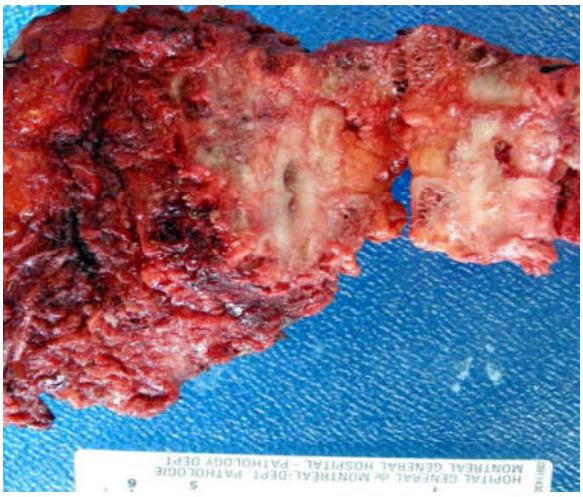

Fig. 7: Right hemivertebrectomy of T11. Examination of the specimen revealed a 1.6 cm liquefied cystic and fibrotic lesion contained in its entirety within the bone. Histologic assessment revealed only fibrohistiocytic change and no evidence of viable sarcoma suggesting a 100% tumor response to pre- operative chemotherapy

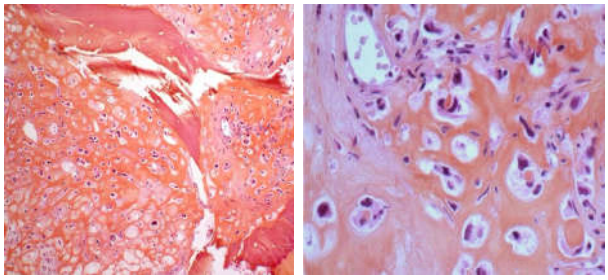

Fig. 8: The cytologic atypia (right) of the malignant cells is severe and is characterized by large irregular cells with large hyperchromatic nuclei and irregular nuclear membranes. The immature bone matrix is directly laid down by the malignant cells. (hematoxylin phloxine saffron stain (HPS), 400 X magnification). High grade Osteoblastic Osteosarcoma (left). Malignant cells produce abundant amount of immature non-mineralized osteoid bone matrix in a lace-like pattern and infiltrate between pre- existing cancellous bone trabeculae. (HPS stain, 100 X magnification)

Postoperative Course

Postoperative course was complicated by cerebrospinal fluid (CSF) leak, which drained into the chest tube for the first 4 days post op, but then sealed spontaneously. Postoperative adjuvant chemotherapy was administered consisting of 6 successful cycles of Adriamycin and Cisplatinum. The patient suffered significant local back pain, which was difficult to manage but eventually controlled medically. The patient is now 42-months post op. She has no neurological deficit and is radiologically free of disease. Recently, she underwent a healthy pregnancy and delivered her first child.

Discussion

Epidemiology

Vertebral osteosarcoma is a rare malignant tumor, that carriesa poor prognosis [5,11,12]. It represents about 3.6% to 14.5% of primary spinal tumors [5,13,14]. The reported incidence of vertebral osteosarcoma ranges between 0.85% and 3% of all osteosarcomas [5,11,15,16].

Dural Resection as Part of Total En Bloc Spondylectomy

It is said that the dura serves as a barrier layer against tumor invasion into the dural sac [1]. From the data available, this appears to be right but there is no strong histological evidence yet. This is likely dueto the fact that total en bloc spondylectomy usually spares the dural sac [7,17]. Tomita accepted intralesional margins if the tumor extendedinto the spinal canal [2]. However, the Weinstein, Boriani, Biagnini (WBB) staging system dictatesto includethe

dura in the en bloc resection whenever the tumor shows an epidural extension [14]. Our case shows that it is possible, at least in some cases, to resect the dura and maintain neurologic integrity.

Our review of the literature revealed few cases where the dura was resected in tumors of the mobile spine (Table 1). One report only was a case of metastatic tumorwith gross dural infiltration but with no microscopic details provided [9]. In most of the other cases (4 out of 7) that were presented with reasonable pathological data regarding dural involvement, the dura was grossly intactin one case [5], had an epidural invasion in another case [18] andwas histologically [7] not invaded in 2 cases, even thoughthe dura was macroscopically destructed in one of them. Our case also showed no microscopical invasion in spite of the suspected dural adhesions to the tumor. This indicates thatdural resection, when done in compliance to oncological principles, should not lead to tumor cell dissemination into the dural space. All 6 alive patients (including ours) had no evidence of local recurrent disease on last follow up ranging between 6-116 months. In two patients [7] who died, the cause of death was metastatic disease. The third deceased patient was reported to have no local recurrence; however, the cause of death was not mentioned [4].

Dural resection in an already high-risk operationshould be only indicated if surgical risks and added neurologic risk are justified to increase survival and wide margins could not be secured otherwise [6]. In our review, the only report with a detailed histopathological analysis described two cases in which a pseudomembrane between the dura and tumor was absent [7]. This may indicate that in cases of dural involvement, even marginalmargins might not be achieved with resection of the dura. We think that including the dura is necessary to achieve wide or marginal margins when there is radiological or intraoperative suspicion of epidural/dural extension of the tumor. One must also consider the risk of violating a natural barrier to tumor expansion and dissemination into the subarachnoid space and CSF. Indeed, Krepler et aladvocated dural resection only if the tumor extended in less than a third of the dural circumference [6]. In aseries of 13 patients the authors reported removing the superficial layer of the dura in cases of epidural tumor extension [19]. There was no more information on the number of cases for which they did dural resection, but they mentioned that this technique was not feasible in the lumbar spine because the dura is too thin. However, there are other reports that described a successful resection of a full dural circumference [5,7]. In all of these reports, the dural resection was part of the resection of both the dura and the spinal cord at the levels of spondylectomy. Biagini described the only case of anterior dural removal, which was technically challenging [10]. Good response to neoadjuvant therapy may help saving the dura from resection [4,6].

Complications

En bloc spondylectomiescarry ahigh risk of complications and should be therefore managed by a multidisciplinary team approach [4,20]. Complications of en bloc resections of spinal tumors need to be studied in more standardized methods [20]. Reported complications include prolonged anaesthesia time, massive bleeding, dural or neurological injuries, wound necrosis, epidural abscesses, sepsis and intraoperative myocardial infarction (MI). In the long term, non-union, hardware failure and chronic dysphagia were reported [1,4,5,8]. Perioperative death occurred in 1 to 5% of the cases across the reported series [8].

Dural resection carries the potential risk of delaying chemotherapy and/or radiotherapy [3]by prolonging the recovery time. It may also cause an intradural spread of tumor cells especially if the inner layer of the dura was infiltrated. Replacement of the dura may also increase the theoretical risk of encephalomyelitis in case of wound infection [6]. Fisher reported a case of subarachnoid pleural fistulae with dural resection. Fisher's case was complicated by pleural effusion and hydrocephalus of undetermined cause [4]. The patient died of metastasis 22 months later.

Conclusion

Total en bloc spondylectomy should be carried out for primary spinal tumors if the benefits outweigh the risks. This complicated procedure must be approached by a multidisciplinary team. We believe that including the dura in the en bloc resection is feasible although it increases the risks of complication in an already complex surgery. Thus, en bloc spondylectomy with dural excision in the setting of primary spine malignancy should only be undertaken when wide margins are unachievable by other means. The dura seems to be an obstacle for intradural tumor invasion but the data in this regard is insufficient.

References

1. Krepler P, Windhager R, Bretschneider W, Toma CD,

Kotz R. Total vertebrectomy for primary malignant tumours of the spine. J BONE Jt Surg. 2002;84-B(5):712-715.

2. Tomita K, Kawahara N, Baba H, Tsuchiya H, Fujita T, Toribatake Y. Total En Bloc Spondylectomy- A New Surgical Technique. Spine (Phila Pa 1976). 1997;22(3): 324-333.
3. Boriani S, Biagini R, De Iure F, et al. En bloc resections of bone tumors of the thoracolumbar spine. A preliminary report on 29 patients. Spine (Phila Pa 1976). 1996;21(16):1927-1931. doi:10.1097/00007632-199608150-00020.
4. Fisher CG, Keynan O, Boyd MC, Dvorak MF. The Surgical Management of Primary Tumors of the Spine Initial Results of an Ongoing Prospective Cohort Study. Spine (Phila Pa 1976). 2005;30(16):1899-1908.
5. Keynan O, Fisher CG, Boyd MC, O'Connell JX, Dvorak MF. Ligation and partial excision of the cauda equina as part of a wide resection of vertebral osteosarcoma: a case report and description of surgical technique. Spine (Phila Pa 1976). 2005;30(4):97-102. doi:10.1097/01.brs.0000153396.39009.a3.
6. Krepler P, Windhager R, Toma CD, Kitz K, Kotz R. Dura resection in combination with en bloc spondylectomy for primary malignant tumors of the spine. Spine (Phila Pa 1976). 2003;28(17):E334-8. doi:10.1097/01.BRS.0000090504.32585.AC.
7. Murakami H, Kawahara N, Tsuchiya H, Demura S, Yamaguchi T, Tomita K. Invasive features of spinal osteosarcoma obtained from whole-mount sections of total en bloc spondylectomy. J Orthop Sci. 2007;12(3):311-315. doi:10.1007/s00776-007-1114-5.
8. Ahn J, Colman MW. Spine Surgery En Bloc Spondylectomy for Primary Review of Reported Outcomes. Contemp spine Surg. 2017;17(4):1-8.
9. Aryan HE, Acosta FL, Ames CP. Two-level total en bloc lumbar spondylectomy with dural resection for metastatic renal cell carcinoma. J Clin Neurosci. 2008;15(1):70-72. doi:10.1016/j.jocn.2006.01.023.
10. Biagini R, Casadei R, Boriani S, et al. En bloc vertebrectomy and dural resection for chordoma: a case report. Spine (Phila Pa 1976). 2003;28(18):E368-72. doi:10.1097/01.BRS.0000084644.84095.10.
11. Barwick KW, Huvos AG, Smith J. Primary osteogenic sarcoma of the vertebral column. A clinicopathologic correlation of ten patients. Cancer. 1980;46(3):595-604. doi:10.1002/1097-0142(19800801)46:3<595::AID-CNCR2820460328>3.0.CO;2-8.
12. Bielack SS, Wulff B, Delling G, et al. Osteosarcoma of the trunk treated by multimodal therapy: Experience of the cooperative osteosarcoma study group (COSS). Med Pediatr Oncol. 1995;24(1):6-12. doi:10.1002/mpo.2950240103.
13. Dreghorn CR, Newman RJ, Hardy GJ, Dickson RA. Primary tumors of the axial skeleton. Experience of the Leeds Regional Bone Tumor Registry. Spine (Phila Pa 1976). 1990;5(12):137-140.
14. Weinstein J, McLain R. Primary tumors of the spine. Spine (Phila Pa 1976). 1987;12(9):843-851.
15. Campanacci M, Cervellati G. Osteosarcoma: A review of 345 cases. Ital J Orthop Traumatol. 1975;1(1):5-22.
16. Huvos AG, Rosen G, Bretsky SS, Butler A. Telangiectatic osteogenic sarcoma: A clinico-pathologic study of 124 patients. Cancer. 1982;159: 1679-1689. doi:10.1002/1097-0142(19820415) 49:8<1679::AID-CNCR2820490824>3.0.CO;2-2.
17. Fujita T, Ueda Y, Kawahara N, Baba H, Tomita K. Local spread of metastatic vertebral tumors. A histologic study. Spine (Phila Pa 1976). 1997;22(16):1905-1912. doi:10.1097/00007632-199708150-00020.
18. Chanplakorn P, Chanplakorn N, Pongtippan A, Jaovisidha S, Laohacharoensombat W. Recurrent epithelioid sarcoma in the thoracic spine successfully treated with multilevel total en bloc spondylectomy. Eur spine journale spine J. 2011;20(SUPPL. 2):S302-S308. doi:10.1007/s00586-011-1723-8.
19. Hasegawa K, Homma T, Hirano T, et al. Margin-Free Spondylectomy for Extended Malignant Spine Tumors Surgical Technique and Outcome of 13 Cases. Spine (Phila Pa 1976). 2007;32(1):142-148.
20. Yamazaki T, Mcloughlin GS, Patel S, Rhines LD, Fourney DR. Feasibility and Safety of En Bloc Resection for Primary Spine Tumors A Systematic Review by the Spine Oncology Study Group. Spine (Phila Pa 1976). 2009;34(22S):S31-S38.