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Case Report

Circumferential Patching of the Dura Mater in the Setting of Dural Fistula after En Bloc Spondylectomy and High Dose Radiation Therapy

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Abstract

A rare neoplasm derived from notochordal remnants, chordoma has a well-characterized tendency to recur following an intralesional resection. Margin-free total en bloc spondylectomy (TES) is the only surgical approach associated with no tumor recurrence at follow-up longer than 5 years. Incidental durotomy is a common complication of TES. We report a novel technique to repair incidental durotomies in two patients who underwent TES for thoracic/thoracolumbar chordoma – a 76-year-old male who presents with an L1 chordoma and a 65-year-old female who presents with acute spinal cord compression from recurrence of a T9-T11 chordoma.

Both patients underwent high dose pre-operative radiation therapy and TES for resection of their respective tumors. Both patients developed incidental durotomies that were initially repaired by primary suture. As we were concerned with the irradiated dura's ability to heal and hold sutures, we supplemented primary repair with collagen matrix grafts. For each patient, the graft was passed in front of the thecal sac and wrapped circumferentially around the thecal sac, forming a seal. After repair, the patients recovered without complications.

Here we describe two patients who presented with chordoma of the thoracic/thoracolumbar spine and have undergone TES for tumor resection. Due to preoperative radiation therapy, this population may have friable dura that are not amenable to repair by primary suture alone in the event of incidental durotomy. We thus augmented our primary repair by passing a collagen matrix graft circumferentially around the thecal sac. These procedures were greatly aided by the maximum accessibility provided by the initial TES procedures.

ABBREVIATIONS

TES: Total en bloc spondylectomy, **CSF**: cerebrospinal fluid, **Gy**: Gray unit, **IMRT**: intensity modulated radiation therapy, **VRE**: vancomycin-resistant enterococci

INTRODUCTION

Surgical management of primary malignant spinal tumors and solitary spinal metastases is often challenging. Taken together with neurological and stability concerns, the anatomical proximity of a primary vertebral tumor to major vessels has made curettage and piecemeal excision a common practice in spinal oncology [1,2]. Such an intralesional approach introduces a high risk of tumor cell contamination of surrounding structures, as it is difficult to differentiate healthy tissue from tumor. This tumor margin violation increases the likelihood of local recurrence of the malignancy [1].

When negative margins are obtained, TES reduces the recurrence rate of spinal malignant tumors.[2] Importantly, TES in patients with primary malignant tumors and spinal metastases has been shown to have superior outcomes to curettage and piecemeal resection [3,4]. We perform this procedure in two stages – a posterior stage and an anterior stage. In the first

stage, we perform a posterior laminectomy with posterior spinal instrumentation. In the second stage, we perform an en bloc corpectomy with anterior spinal fusion and instrumentation.

Here we discuss two patients who presented with chordomas of the thoracic and thoracolumbar spine. Commonly occurring in the skull base and vertebral column, chordomas have a well-characterized tendency to recur following intralesional resection [5,6]. Indeed, uncontrolled growth of this tumor after surgery is a common cause of death in patients with chordoma. A review of the largest series of chordoma in the mobile spine has shown that margin-free TES is the only treatment associated with no recurrence at follow-up longer than 5 years [5].

A common complication of TES is incidental durotomy [6]. If left unmanaged, an incidental durotomy with concomitant CSF leakage may result in adverse sequelae such as sepsis and meningocele [7]. The current report describes a novel technique to repair incidental durotomies in two patients treated with high dose pre-operative radiation therapy who underwent TES for chordoma of the thoracic and thoracolumbar spine. To our knowledge, such an approach has not been previously reported.

CASE PRESENTATION

Patient 1 is a 76-year-old male with a chordoma of the L1 vertebral body with associated severe compression fracture. He received high dose pre-operative radiation therapy: 50.4 Gy in 28 fractions (19.8 Gy IMRT with 6 MeV photons, 30.6 Gy pencil-beam scanning protons) over 41 elapsed days.One month after completing his pre-operative radiation regimen, the patient underwent a two-stage en bloc spondylectomy for the thoracolumbar chordoma; the first stage consisted of a posterior laminectomy with fusion and the second stage consisted of an anterior corpectomy with reconstruction. The L1 vertebra was completely circumferentially resected. He also underwent a partial left nephrectomy for renal cell carcinoma with clear margins. We were unable to perform intraoperative dural plaque brachytherapy due to anatomic constraints that prohibited the safe administration of radiation. After recovery, the patient displayed altered mental status. We then obtained a CT scan, which showed a possible hygroma. This finding led to a subsequent positive CT myelogram that was consistent with the patient's altered mental status and possible hygroma.

We thus decided to explore the thoracolumbar operative site and inspect the dura. The patient was placed prone and then in the Trendelenburg position, with anesthesia induced intravenously through his tracheostomy tube. The patient was maintained in the Trendelenburg position after general endotracheal anesthesia was induced. After appropriate preparation and draping, we approached the spinal cord using our previously placed posterior midline incision. From the incision, we removed the posterior rods to gain access to the dura. We obtained a clear view of the dura after removing the rods. The thecal sac had very good turgor. While there was copious clear fluid, we did not observe fluid directly leaking from the dura. No dural leak was noted ventrally upon examination.

Although no direct leakage of CSF was observed, we noticed a tear in the axilla of the T12 nerve root on the left side. The arachnoid layer was intact and no active CSF leakage was observed. The T12 nerve root was under tension. We ligated the root approximately 3 centimeters from its source out of the thecal sac. We then used the nerve root as a graft and sutured it to the thecal sac, thereby closing and grafting the tear found in the axilla.

Concerned that there may have been an occult dural injury that was not appreciable from our posterior approach, we elected to wrap a collagen matrix graft (DuraMatrix from Stryker Corporation, Kalamazoo MI) circumferentially around the thecal sac (Figure 1). The graft was passed in front of the thecal sac behind the expendable cage and wrapped 360 degrees around the thecal sac. Prior to suturing the dural graft in place, we covered the graft in a fibrin sealant (Tisseel from Baxter International

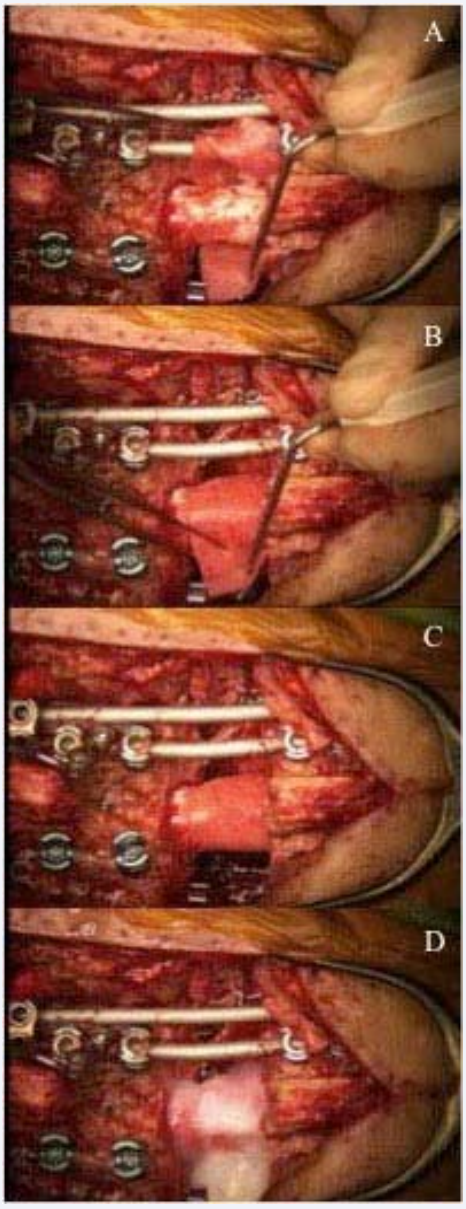

Figure 1 The Dura Matrix was passed in front of the thecal sac (A), wrapped circumferentially around the thecal sac (B), and sutured in place (C). The remaining wound and graft were covered with Tisseel (D). The graft was not sutured directly to the dura.

Inc., Deerfield IL). The dural graft was sutured in place with two synthetic sutures; the graft was not sutured directly to the dura. The remaining wound and graft were covered with Tisseel, using 20 mL total of Tisseel. We closed the wound in layers, closing the dead space. No drain was used. Excellent hemostasis was obtained.

Post-repair, the patient suffered from 10 days of delirium and fever. He was treated for VRE surgical bed and hardware infection, VRE meningitis, and Pseudomonas urinary tract infection. After prolonged antibiotic treatment, the patient recovered from these infections and has made good progress undergoing rehabilitation. His wounds healed completely. Six months after tumor resection, he received a post-operative radiation boost encompassing the tumor bed and immediately adjacent tissue: 21.6 Gy (pencil-beam scanning protons) in 12 fractions over 18 elapsed days. He has had no tumor recurrence to date.

Patient 2 is a 65-year-old female who presented with acute spinal cord compression from local recurrence of a T9-T11 chordoma. She had previously undergone a posterior stabilization procedure with placement of hardware and placement of thread wire saws (Medtronic, Minneapolis MN) about the thoracic spine. She was felt to be a candidate for a total en bloc spondylectomy of T9, T10, and T11. The patient received pre-operative high-dose adjuvant radiation therapy: 50.4 Gy in 28 fractions (19.8 Gy IMRT with 6 MeV photons, 30.6 Gy three-dimensional conformal protons) over 36 elapsed days. One month after completing her pre-operative radiation regimen, she underwent TES. We administered intraoperative dural plaque brachytherapy (10Gy) about the T9-T11 dura to boost the dural surface during surgery.

For the TES, she was placed in a lateral decubitus position and her thoracic spine was exposed through a left-sided thoracotomy. After the thoracic spine exposure, we identified the thread wire saws that had been passed in the previous surgery and sutured to the rods from the stabilization on the left side. We cut the sutures that were holding the thread wire saws to the rods. Applying the handle to the thread wire saw, we cut the vertebral body in a sawing motion between T8 and T9 at the caudal aspect of T8 and again at the cephalad aspect of T12. The tumor was excised and its margins were grossly negative.

The tumor was gently rotated and adhesions to the dura were removed. We encountered lateral and dorsal durotomies with CSF leakage that were repaired primarily with sutures. We then elected to pass a collagen matrix graft (DuraGen from Integra LifeSciences Corporation, Plainsboro NJ) circumferentially around the thecal sac to augment the primary repair (Figure 2). We sized the DuraGen patch so that it would span the 3-segment defect left after the corpectomies. The ends of the DuraGen patch were sutured, thereby effecting a seal. At this point, the entire dura had been completely circumferentially covered. The DuraGen graft was not directly sutured onto the dura.

Anterior spinal instrumentation with fusion of T8-L1, anterior spinal instrumentation with femoral ring allograft of T12-L1, and anterior spinal instrumentation with a large 3-segment titanium mesh cage spanning from T8-T12 were performed to complete the case. The wound was closed in layers.

Two months after tumor resection, the patient received post-operative radiation encompassing the tumor bed and immediately adjacent tissues: 19.2 Gy (three-dimensional conformal protons) in 12 fractions over 19 elapsed days. She has made excellent progress in physical therapy and is gradually returning to her previous level of function. She has had no tumor recurrence to date.

Figure 2 The Dura Gen was passed circumferentially around the thecal sac (A, B) and wrapped circumferentially around the thecal sac (C). The ends of the DuraGen patch were sutured (D). The graft was not sutured directly to the dura.

DISCUSSION

Incidental durotomy is a relatively common complication of spinal surgery, occurring in 1% to 5% of spinal surgeries [7-10]. The incidence of dural tears increases with the complexity of surgery [7,8]. Indeed, dural tears are common complications of highly complex TES operations [6]. When associated with

CSF leakage, an incidental durotomy may introduce the risk of pseudomeningocele formation, subdural hematoma, hygroma, and postoperative infection [11-15]. Complete primary repair is thus recommended in the case of a dural tear, regardless of the tear size [11,16].

Dural repair techniques aim to contain nerve roots and achieve closure of the dural sac without inducing a significant local inflammatory response. Simple durotomies are typically repaired with sutures. CSF leakage after suture repair is significantly decreased with the use of sealants [16,17]. Grafts of muscle, fat, or fascia may be used in addition to fibrin sealant to effectively augment primary suture repair.[8,18] In addition, type I collagen matrix (e.g. DuraGen, DuraMatrix) has been successfully used as a dural graft, as type I collagen is chemotactic to fibroblasts and enhances dural regeneration [19]. An important consideration in patients with incidental durotomies is whether they have undergone radiation therapy. In our clinical experience, irradiated dura does not behave as normal dura with regards to its ability to heal and to hold sutures. Irradiation greatly delays dural healing and increases the risk of dural scarring [13,20]. Primary suture of an irradiated scarred dura is thus less likely to prevent CSF leakage from a durotomy, requiring additional closure superficial to the dura [20].

Here we discuss two patients who presented with chordoma of the mobile spine – one with an L1 and the other with a tumor spanning T9-T11. They were both treated pre-operatively with high dose radiation therapy. For thoracolumbar lesions, we recommend neoadjuvant pre-operative radiation therapy of 50.4 Gyto be followed by post-operative boost radiation to the encompassed tumor bed based on our published research [21,22]. This strategy allows for smaller radiation volumes than exclusive post-operative radiation therapy. This is an important consideration as the spinal cord is sensitive to radiation greater than 45-50 Gy [21,22]. Furthermore, high-dose pre-operative radiation reduces the risk of iatrogenic tumor seeding into the surgical bed [21,22]. The initial pre-operative dose is delivered to the gross tumor as seen on MRI. The vertebrae above and below are also included as well as a margin of soft tissue $\geq$1 cm outside the gross tumor. We utilized intraoperative dural plaques to allow for the delivery of the prescription radiation dose to tumors that abut the dura [21,22]. Resection is usually performed 4-5 weeks after the completion of pre-operative radiation. Post-operative radiation is administered upon sufficient healing of the incision.

Both patients underwent TES with either post-operative (Patient 1) or intraoperative dural CSF leaks (Patient 2). Given the friable nature of the patients' respective dura – likely a result of radiation therapy – we elected to augment our primary repair of the observed durotomies. We employed a circumferential technique in which a collagen matrix graft was wrapped 360 degrees around the thecal sac, achieving complete coverage and effecting a seal. No CSF leaks were observed after repair, and both patients recovered without complications.

The efficacy of this repair approach is contingent on the accessibility of the thecal sac. Without the posterior laminectomy and en bloc corpectomy of TES, full visualization and manipulation of the thecal sac would not be possible; passing a collagen matrix graft around the thecal sac would have been exceedingly difficult. We were thus aided by the circumferential resection of the patients' respective tumors.

We have described an effective repair technique for incidental durotomies encountered in a specific population: patients who present with chordoma of the thoracic/thoracolumbar spine, have been treated with preoperative high dose radiation therapy, and have undergone TES for tumor resection. Repair by primary suture alone may not be adequate in the event of an incidental durotomy, as the patient's dura may be irradiated and scarred. We thus describe a technique to pass a collagen matrix graft circumferentially around the thecal sac to supplement primary repair. This approach is greatly aided by the maximum accessibility to the thecal sac provided by the previously performed TES.

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