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Sacral insufficiency fractures are a risk of massive bleeding during sacrectomy: patient series

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BACKGROUND Sacrectomy carries significant risk of bleeding; however, specific risk factors, apart from medical comorbidities and tumor type, for this life-threatening complication remain unclear. This study describes two cases of massive bleeding, including one death during sacrectomy attributable to adherence of the internal iliac vein (IIV) and its neuroforaminal tributaries from sacral insufficiency fractures.

OBSERVATIONS The authors presented two cases involving patients who received sacrectomy for a chordoma and experienced massive bleeding from the IIV due to adherence of the IIV and its neuroforaminal tributaries around sacral insufficiency fractures. They assessed their institution's previous two decades' experience of sacrectomies to determine risk factors for massive bleeding and performed anatomical dissection of 20 hemipelvises, which revealed the close proximity of the IIV to the sacral foraminae and the consistency of neuroforaminal tributaries arising from the foraminae.

LESSONS Sacral insufficiency fractures may cause scarring that adheres to the IIV and its neuroforaminal tributaries, which risks massive bleeding during sacrectomy.

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KEYWORDS spine; tumor; cancer; bleed; transfusion

Sacral resection is an uncommon procedure with significant risks, including massive life-threatening hemorrhage.[1,2] However, this procedure is reserved for patients with either curative malignant disease or decompression of compressive noncurative lesions to improve functional outcome.[1] Thus, in some patients the risks are outweighed by the potential benefits.

In such patients, preoperative planning is critical to achieve an adequate resection, optimize functional outcomes, and avoid complications. Although some complications cannot be predicted or avoided, the avoidance of massive life-threatening bleeding is always considered in these cases. Techniques to avoid this complication include preoperative embolization, use of perioperative procoagulants, dual discipline surgery, combined anterior and posterior approaches, and careful dissection.[3–6]

Most bleeding is experienced from the pelvic venous plexus or from the lesion itself, however, depending on the lesion type.[7] This bleeding is typically a constant leak that requires mechanical compression while the lesion is excised. Once clearly visible, it is treated with systematic ligation of each bleeding source. Although a large volume of blood can be lost from this type of bleeding, the blood loss can usually be compensated for with reinfusion or transfusion. In contrast, massive life-threatening hemorrhage typically results from a major vessel bleed and occurs rapidly, presenting significant challenge in maintaining hemodynamic control.[7]

The perisacral anatomy is complex and often variable; however, the major vessels likely to cause life-threatening hemorrhage during sacrectomy are the internal iliac vein (IIV) and internal iliac artery.[8,9] These vessels lie close to the anterior sacrum and are bound to

ABBREVIATIONS EBL = estimated blood loss; IIV = internal iliac vein.

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the sacrum by small tributaries that perforate the anterior foraminae. However, in the nonpathological state, these vessels remain mobile and are easily manipulated on the foraminal perforating vascular pedicles. In contrast, in the pathological state, either from direct lesion infiltration or postradiotherapy scarring, these vessels can adhere to the tumor mass and make dissection difficult.[7]

In the pathological sate, fractures of the sacrum can also occur; however, the implications of such events remain unclear. This study presents two rare cases of massive intraoperative bleeding during sacrectomy that was attributed to adherence of the IIV and its neuroforaminal tributaries to the sacral insufficiency fractures. We analyzed the experience of massive bleeding during these procedures at our institution over the last 2 decades. We also dissected 20 hemipelvises to anatomically define the proximity of the iliac vessels to the sacral foraminae.

Study Description

We presented two patients with sacral insufficiency fractures receiving sacrectomy for a chordoma resection who experienced massive bleeding due to adherence of the IIV and tributaries to the fractured sacrum.

We subsequently reviewed the previous 24 consecutive adult sacrectomies performed within our institution between 2000 and 2019 with a minimum 2-year follow-up, specifically assessing for the number of patients with massive intraoperative hemorrhage that was in excess of 5 L or necessitated a massive transfusion protocol, and we assessed the cause for such hemorrhages. Patient age, sex, tumor type, surgical approach, estimated blood loss (EBL), and surgical complications were reviewed.

We then performed anatomical dissection of 20 cadaveric fresh hemipelvises without previous abdominal or pelvic surgery or macroscopically identifiable abdomino-pelvic pathology to assess proximity of the IIV to the sacrum and its anatomical relationships (University of Otago Institutional Review Board ethical approval). Dissection was performed through an anterior approach with resection of the abdominal musculature and gastrointestinal organs to expose the aorta and inferior vena cava. Loupe magnification was used to dissect the vessels into the pelvis, preserving their anatomical position and tributaries to the sacrum. We assessed the height above (+) or below (−) the sacral promontory where the common iliac vein bifurcated and termed this the height separation. We then measured the length and diameter of the common iliac vein. Subsequently, we measured the distance of the IIV lateral of the median sagittal line at the given level of each neuroforamen, termed the S1–S3 median. Finally, we measured the distance of the IIV from the medial border of respective neuroforamen, termed the S1–S3 foramen, which represents the length of the tributary veins. All measurements were performed with calipers. The medial border of the sacral foramen was used because it offered a sharp, easily defined edge in contrast to the foramen's lateral border, which was seen to be smooth and rounded for the contour of the exiting nerve root but did not permit a clearly definable border.

Statistical Analysis

For the cadaveric analysis, we used a Kolmogorov-Smirnov test for normality assessment. After this analysis, an ordinary one-way analysis of variance was deployed with a p value set at $\leq$0.05.

Illustrative Cases

Case 1

A 61-year-old previously healthy man presented with bilateral lower limb neurological dysfunction and pain from a massive sacral chordoma. Preoperatively, he had not had radiotherapy or previous operative intervention, but he did have tumor-related bilateral sacral insufficiency fractures (Fig. 1).

He received a sacrectomy through a posterior midline incision and Mercedes-Benz extension. Initially, the L3–L5 spine was exposed to allow insertion of bilateral pedicle screws into L3 and L4 under fluoroscopic guidance. The gluteus maximus was then elevated to the sciatic notch, followed by incision of the sacrospinous/sacrotuberous ligament complex. The superior gluteal nerve and artery as well as the sciatic nerve were identified and protected.

The anterior sacrum was freed with a digit bilaterally from the coccyx to the S3 and then from S1 to the sciatic notch just lateral to the sacroiliac joints for later osteotomy. A bilateral L5 laminectomy and inferior articular process resection was then performed. The dura and neural elements caudal to the L5 nerve roots were divided and a watertight suture repair was performed. The plane anterior to the L5 body was then developed and the L5 nerve roots partially mobilized. The L5 vertebral body was split in the axial plane with a combination of an ultrasonic bone scalpel and Kerrison rongeurs, with careful protection of the anterior structures. The iliac osteotomies were then completed.

Gradual mobilization of the sacrum/tumor was performed, but the L5 nerve roots and IIV were seen to be highly adherent to the sacrum. During mobilization of the right L5 nerve root from the sacrum, significant venous bleeding was encountered from the IIV and its neuroforaminal tributaries, which was controlled with packing. We proceeded with freeing of the left sacral alar, but once again significant venous bleeding was encountered from the IIV and its neuroforaminal tributaries while trying to free the anterior structures from the lateral sacrum. Unlike the right side, the bleeding was uncontrollable with packing and compression. Attempted visualization of the bleeding vessels failed.

In a lifesaving attempt to visualize and control the bleeding, we elected to rapidly deliver the sacrum/tumor and directly compress the bleeders. At this stage, it was clear that the IIV had been torn bilaterally due to marked adherence of its neuroforaminal tributaries to the anterior sacrum in the region of the previous sacral insufficiency fractures. There was no adherence of the tumor itself to these vessels. Rapid packing and closure were performed before the patient was placed supine and a laparotomy with direct inferior vena cava and aortic compression was performed. Although this approach allowed improved control of the bleeding, the patient experienced cardiac arrest from hypovolemia despite a massive transfusion protocol (24 units of red blood cells, 10 units of fresh frozen plasma, 4 units of platelets, and 4 units of cryoprecipitate) and died intraoperatively from extensive bleeding.

Case 2

A 69-year-old previously healthy man presented with low back pain after falling down stairs. He had no bowel or urinary symptoms and was comfortable with analgesia with some mild S1 impairment in the right foot. MRI showed that he had large sacral chordoma extending into the presacral space and the right gluteal region and

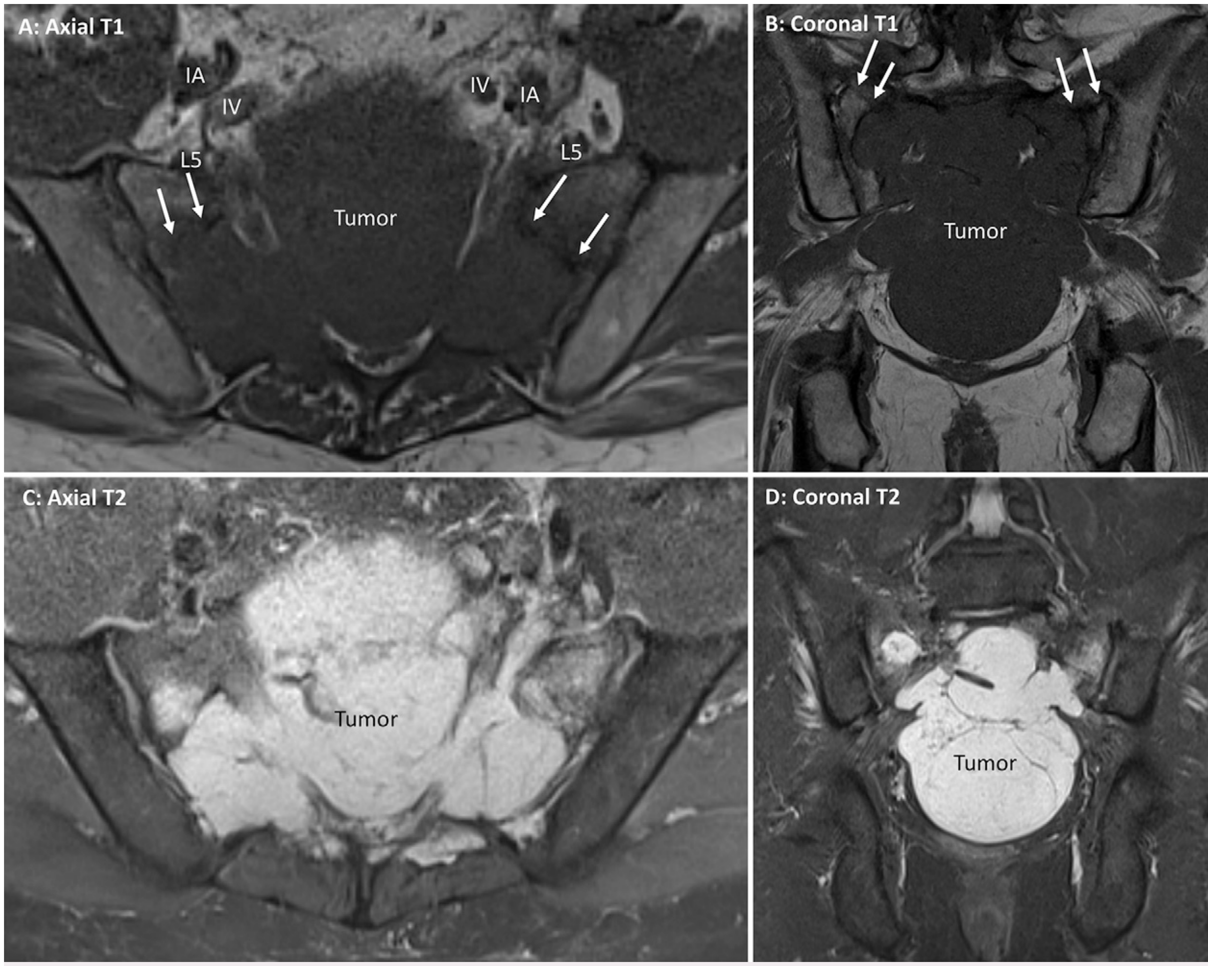

FIG. 1. MRI of the tumor (labeled) and the sacral insufficiency fracture (*arrows*). The iliac artery (IA), iliac vein (IV), and L5 nerve root (L5) are labeled. **A:** Axial T1. **B:** Coronal T1. **C:** Axial T2. **D:** Coronal T2.

crossing the right sacroiliac joint to involve the right ilium (Fig. 2). A CT scan revealed a pathological fracture of the right sacral alar in close proximity to the internal iliac vein (Fig. 2).

He then received a three-stage resection and reconstruction starting with end colostomy and anterior mobilization of the tumor, completion of the osteotomy and resection of the tumor via a posterior approach to the lumbopelvic junction, and finally reconstruction with a vascularized fibula and latissimus dorsi free flap.

The first stage was completed without concern; however, because the tumor mass obscured visualization of the right IIV and its neuroforaminal tributaries, the IIV was not completely mobilized from the sacrum, with the intention of posterior ligation during delivery of the tumor.

During the second stage, posterior soft tissue mobilization, including ligation of the involved right sciatic nerve, iliac bone cuts, L5/S1 discectomy, alar soft tissue resection, and bowel resection were completed. However, while performing the final tumor mobilization from the posterior approach before delivery of the tumor, marked adherence of the right IIV and neuroforaminal tributaries was encountered, with an injury to the IIV resulting in massive intraoperative blood loss of 15 L followed by hypovolemic cardiac arrest. Successful resuscitation with 27 units of red blood cells, 12 units of fresh frozen plasma, 2 units of platelets, and 4 units of cryoprecipitate was performed. After packing, partial but inadequate vascular control was achieved and the decision was made to undertake rapid removal of the remainder of the tumor attachment, including the left sciatic nerve, for speed of delivery. Adherence of the right IIV and its neuroforaminal tributaries was seen in the region of the pathological sacral fracture; no adherence of the tumor itself to the vein was seen. The injury was considered the result of avulsion off the IIV of the adhered neuroforaminal tributaries. This was repaired, and ligation of multiple minor bleeding points was undertaken. Hemostasis was achieved and the patient stabilized. Instrumentation was then performed.

The following day, the third stage of bone and soft tissue reconstruction was performed with a vascularized fibular and a free latissimus dorsi flap. The patient stayed in the intensive care unit for 16 days.

At 3 years after surgery, a small local recurrence was identified, and the patient received additional resection followed up with active surveillance. Postoperatively, he remains disease-free at 8 years. He uses a wheelchair but can stand to transfer.

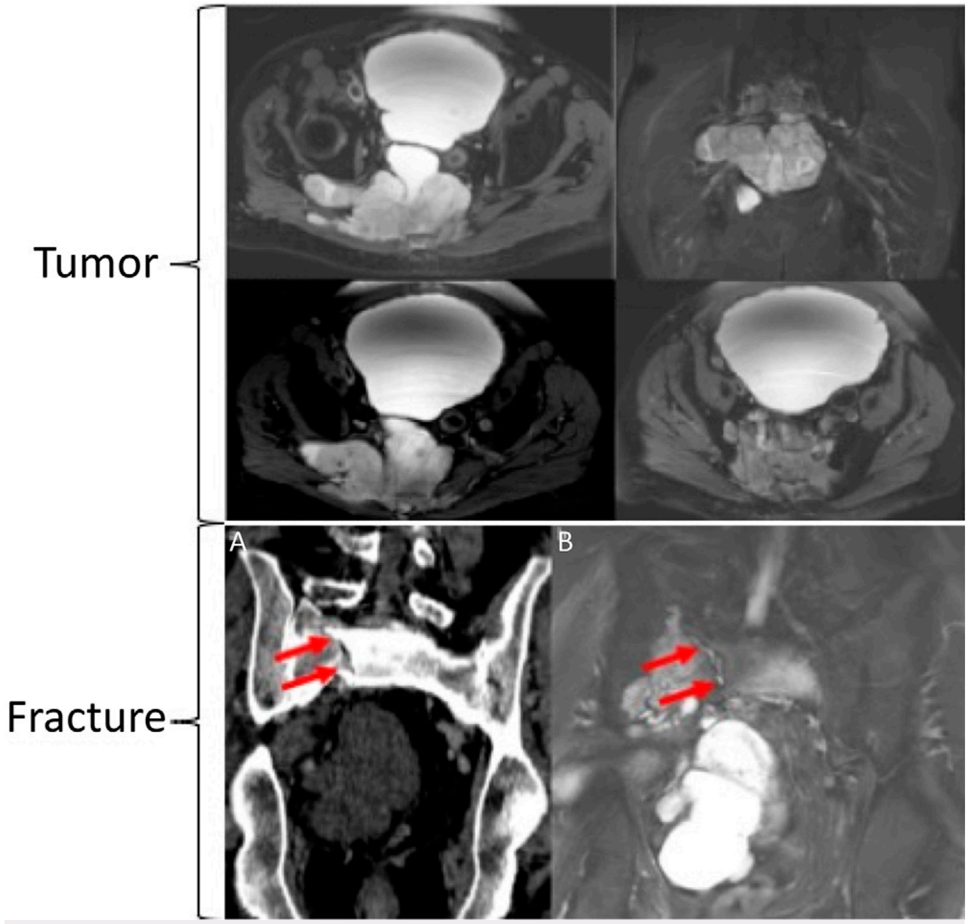

FIG. 2. Preoperative MRI showing extent of tumor (labeled) involvement, including right sciatic notch, both sacroiliac joints, and the posterior aspect of the right hip (**top photo**). CT (**A**) and MRI (**B**) showing preoperative pathological fracture (*arrows*).

Review of Previous Cases

Table 1 summarizes the previous 24 consecutive cases, excluding the cases described in the case report.

No other cases were identified in this cohort with sacral insufficiency fractures and one other case of direct adherence of the IIV and its tributaries to the anterior sacrum. This case involved a 41-year-old man with a malignant peripheral nerve sheath tumor who underwent a sacrectomy and lumbopelvic reconstruction using a dual anterior and posterior approach. He had tumor adherence to the IIV and its neuroforaminal tributaries that was attempted to be freed during the anterior approach, but it resulted in a venous tear that was repaired but resulted in a massive transfusion for an unreported EBL.

One other patient had a total EBL of 10,000 mL, but that patient had an infiltrative chondrosarcoma undergoing a three-stage procedure involving a colostomy, tumor resection, and flap reconstruction with a vascularized fibula graft. The patient's EBL accumulated over the three stages and never required a massive transfusion, and the patient is now disease-free at 3 years follow-up.

Anatomical Dissection

We dissected 14 male and 6 female cadaveric hemipelvises with a mean age of 80.5 years (standard deviation ±10.8 years) and an even split between left and right sides. The results of the measured variables are shown in Figure 3. No statistical difference was found for sex or side for any of the variables assessed.

A neuroforaminal venous tributary to the IIV was identified at each anterior foramen, the length of which equaled the length of the measured S1, S2, and S3 foramen lengths. Figure 4 illustrates a venous neuroforaminal tributary.

Discussion

Observations

The two reported cases represent the first reported cases of massive hemorrhage encountered during sacrectomy attributed to sacral insufficiency fractures. We believe that the scar tissue caused by these fractures adheres to the IIV and its neuroforaminal tributary vessels arising from the anterior sacral foramina. Thus, during our surgical dissection and release of the anterior sacrum,

TABLE 1. Summary of the previous 24 consecutive cases, excluding the case report

	Value
Age (yrs)	Mean 53 (range 21–74)
Sex	15M, 9F
Tumor type	Primary
	Benign
	Schwannoma 1
	Chordoma 5
	Osteochondroma 1
	Giant cell tumor 1
	Malignant
	Sarcoma 6
	Malignant peripheral nerve sheath tumor 5
	Metastatic
	Leimyosarcoma 1
	Undefined 6
Approach	Ant 2
	Pst 6
	Combined 16
EBL (mL)	Mean 2,600 (range 400–10,000)
Complications	Infection 7
	Wound dehiscence 3
	Acute kidney injury 2
	Metalware failure 2
	VTE 1
LOS (days)	Mean 22 (range 10–40)
Recurrence	1
Metastasis	1
Mortality	1 yr: 0
	2 yr: 2

Ant = anterior; Pst = posterior.
Neurological deficits are not included because of the nature of this procedure requiring variable sacrifice of sacral nerve roots.

these adherent vessels, which can usually be mobilized before ligation, were avulsed from the IIVs, resulting in massive hemorrhage.

In a review of our institution's previous experiences, no other patients were identified as having sacral insufficiency fractures to which we could compare their intraoperative complications. However, one case of direct tumor adherence to the IIV and its neuroforaminal tributaries also resulted in massive blood loss. No other cases of tumor adherence to these vessels were identified, suggesting that the pathophysiological risk of massive bleeding is adherence of the vessels to the anterior sacrum.

In our anatomical dissection analysis of nonpathological pelvises, bifurcation of the common iliac vein typically occurred at the level of the sacral promontory, and the IIV was seen to progressively move more laterally as it descends into the pelvis, making it closest to the S1 sacral foramen (2 mm lateral to the foramen's medial border). However, at the level of all sacral anterior foraminae, the IIV was located within 4 mm of the foramen's medial aspect. This vessel was seen to be held to the sacrum by perforating vessels that

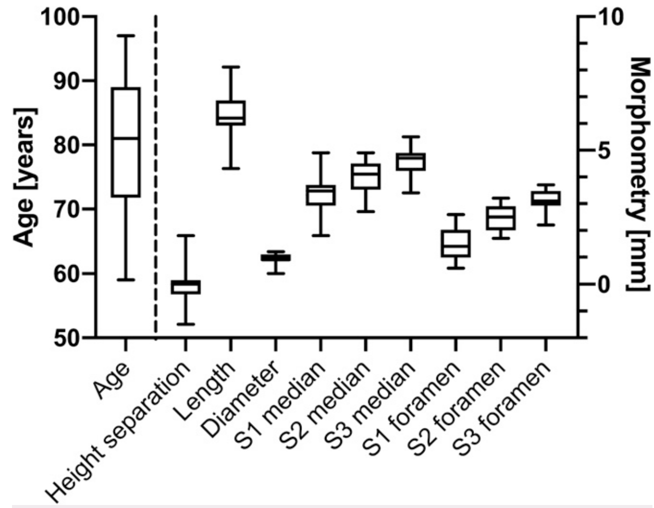

FIG. 3. Summary of the measured variables in the cadaveric analysis.

penetrate each foramen, but these tributaries were all mobile and dissectable in these nonpathological pelvises. This finding confirms that adherence of the vessels to the anterior sacrum is pathological and consistent with the risk of tributary avulsion from the IIV during sacrectomy.

This study has clear limitations. First, these are rare cases of intraoperative massive blood loss during sacrectomy, negating comparative studies. Second, dedicated postmortem dissection was not performed to validate our intraoperative findings. However, despite this difference, we believe that our intraoperative assessment

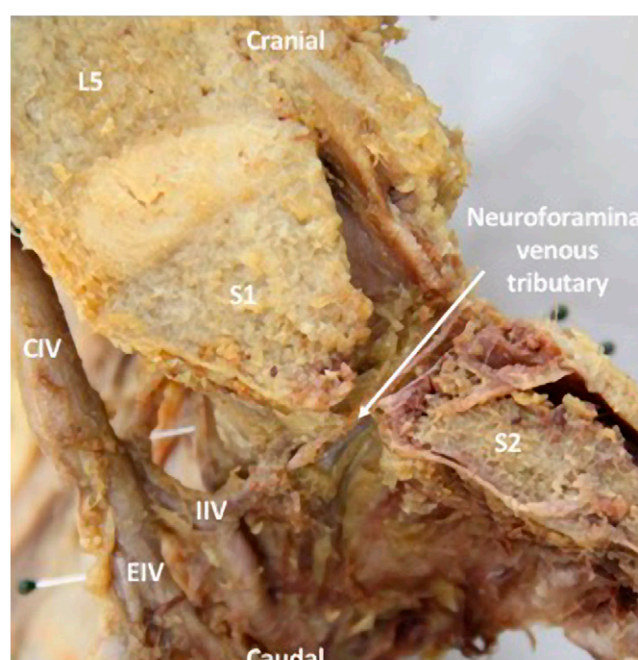

FIG. 4. Sagittal section through the S1 foramen. The common iliac vein (CIV), external iliac vein (EIV), internal iliac vein (IIV), and L5 and S1 vertebrae are labeled.

confirmed vessel adherence to the region of the insufficiency fractures that was not due to direct tumor adherence or any other cause. Third, the review of our institution's experience relied on a retrospective notes review, which limited the data available for extraction. Lastly, our cadaveric analysis was based on nonpathological pelvises, thus not permitting an assessment of vessel position or adherence to the anterior sacrum in the pathological state.

Despite these limitations, our experience suggests that sacral insufficiency fractures cause adherence of the IIV and neuroforaminal tributaries, which increases the risk of massive bleeding during sacrectomy. In such circumstances, we advocate an anterior approach with complete freeing of the IIV and control of its neuroforaminal tributaries before posterior resection and removal of the sacrum.

Lessons

Sacral insufficiency fractures represent a risk of adherence of the IIV and its neuroforaminal tributaries that should be recognized as a risk of massive bleeding during sacrectomy; therefore, preoperative and intraoperative planning should be undertaken.

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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: Kieser, Hammer, Reynolds. Acquisition of data: Kieser, Soltani, Koutp, Reynolds. Analysis and interpretation of data: all authors. Drafting the article: Kieser, Hammer, Hughes, Reynolds. Critically revising the article: Kieser, Hammer. Reviewed submitted version of manuscript: Kieser, Soltani. Approved the final version of the manuscript on behalf of all authors: Kieser. Statistical analysis: Kieser. Administrative/technical/material support: Kieser, Soltani, Hughes. Study supervision: Kieser, Reynolds.

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