CASE REPORT

Challenges of enbloc mid-sacrectomy as redo surgery for sacral chordoma: a case report

Sundus Ali, Ahtsham Ishaq, Adnan Qasim, Muhammad Naeem, Muhammad Rizwan Sarwar, Shahzad Shams

Abstract

Enbloc Sacrectomy is the procedure of choice for aggressive sacral lesions but not widely practiced in Pakistan, both by Neurosurgeons and Orthopaedic surgeons. Only one case has been mentioned in indexed local literature so far and that too not operated in Pakistan. The case of a 27 year old neurologically intact male is presented. He had a huge residual mass and midline non-healing wound after two attempts at intralesional debulking and one full course of local irradiation. He presented to the Mayo Hospital, Lahore on 29th December 2021 for a redo surgery of sacral chordoma. A marginal excision was achieved utilizing posterior only approach. This case will help to understand the key steps in enbloc mid-Sacrectomy and importance of involving multidisciplinary team for ensuring adequate wound closure.

Keywords: Sacral chordoma, Mid-Sacrectomy, Enbloc.

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Introduction

Sacral tumours are rare, require multidisciplinary expertise and have substantial risk of morbidity, therefore surgical experience among local Neurosurgeons and Orthopaedic surgeons is limited [1]. The technical steps for enbloc mid-sacrectomy for chordoma are not dealt so far in any indexed literature in Pakistan. Only one case series mentioned sacral tumours surgery but mainly focussed on lumbo-iliac fixation and not surgical technique of tumour resection [2] This case, therefore attempts to describe the surgical steps for enbloc mid sacrectomy and challenges of a redo sacral chordoma for spinal surgeons.

Case Report

A 27-year-old male was presented in the out-patient department of Mayo Hospital, Lahore on 29 December 2021 with a non-healing wound in mid sacral region (Fig 1a) following intralesional debulking of underlying sacral mass twice, at intervals of 1 year and 4 months before this presentation. He also underwent local irradiation 8 months earlier, as the histopathology was consistent with chordoma. On neurological examination, the patient had paraesthesia in the right buttock, S1-S2 dermatomes and mild sphincter dysfunction but no lower extremity motor weakness. The radiological evaluation of the pelvis via computed tomographic scan (CT) and magnetic resonance imaging (MRI) was consistent with chordoma (Fig1b-d). Single stage enbloc mid sacrectomy was planned via posterior only approach utilizing modified Kraske technique. General anaesthesia was used and the patient was placed in a prone position and a posterior midline incision incorporating 1cm skin margins of previous wound was made (Fig 2a). Bilateral musculocutaneous flaps were raised, leaving a cuff of muscle over the tumour capsule more on right side where the tumour was infiltrating the gluteus muscles (Fig 2b). Anococcygeal ligament was divided and the tip of the coccyx was dissected all around. Proceeding above on either side sacrospinous and sacrotuberous ligaments were identified and divided thus freeing the lower sacrum and allowing gloved hand to be passed in pre-sacral area. The sciatic nerve in the sciatic notch was identified and

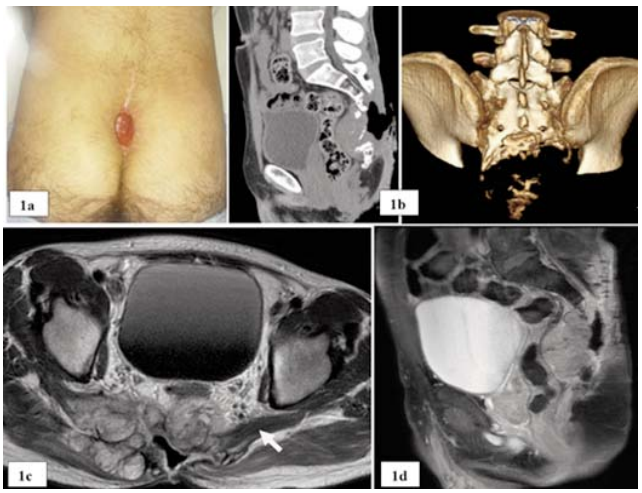

Figure-1: 1a. non healing wound at presentation, 1b CT showing osteolytic lesion involving S3 onwards, 1c Axial MRI depicting left piriformis (white arrow), right one is infiltrated by the lesion, 1d Sagittal T2 to appreciate characteristic hyperintensity of lesion (a known radiological feature of chordoma).

Department of Neurosurgery, King Edward Medical University, Mayo Hospital, Lahore, Pakistan.

Correspondence: Sundus Ali. Email: sundusunn@gmail.com

ORCID ID. 0000-0002-4160-8317

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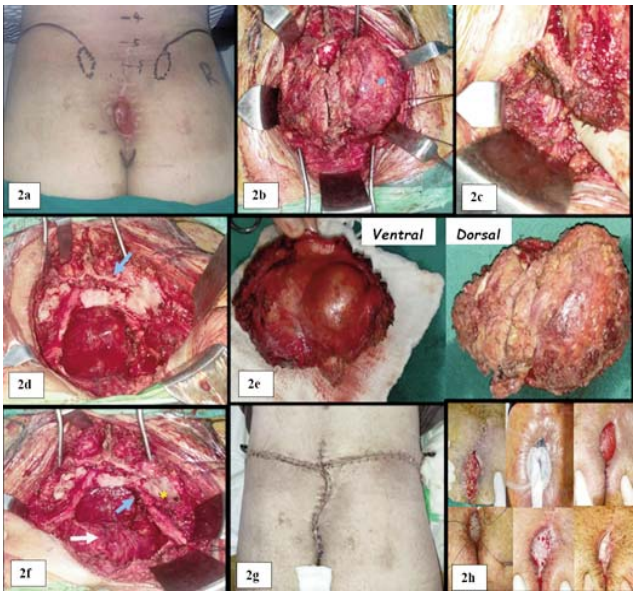

Figure-2: 2a preop marking, 2b cuff of right gluteus maximus over tumour(blue asterisk), dorsal sacral laminectomy (white asterisk) 2c Sciatic nerve in sciatic notch 2d Nerve roots ligated in canal (blue arrow)I, 2e Enbloc specimen, 2f Final view of surgical cavity, Rt. SI joint (yellow asterisk), serosal tears in posterior rectal wall (short white arrows) ligated iliac vessels anterior to Rt SIJ (blue arrow), 2g Bilateral Gluteal rotation flaps, 2h postop Wound dehiscence, VAC and SSG.

preserved on both sides right below the piriformis (Fig 2c). On right side, involved part of piriformis was included. Lowest intact sacral spinous process was confirmed to be S2 after dorsal laminectomy of S3 was done. Four nerve roots were identified inside the canal at the level of laminectomy, out of which three were incorporated into tumour mass, thus ligated, and divided

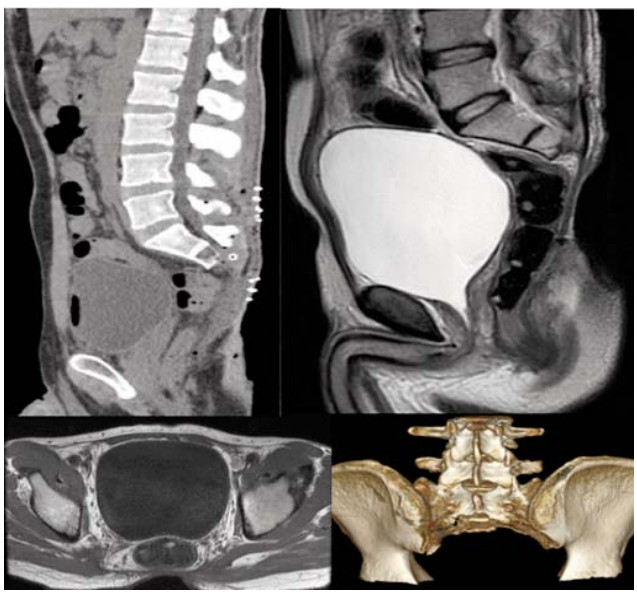

Figure-3: immediate postop CT and 6 weeks MRI showing complete resection.

(Fig 2d). The extreme left root abutting the left lateral border was easily separated from tumour capsule in the last stage.

Finally, ventral bony cuts were made with hammer and chisel guided by the fingertip beneath, thus completing the ventral osteotomy. Right sided sacroiliac joint (SI) was partially involved, and thus lateral osteotomy included part of the joint. As the whole specimen was taken out enbloc (Fig 2e), just ventral to the SI joint, two tributaries of the internal iliac were torn and had to be ligated. The general surgeon on board now inspected the posterior surface of rectum and found two serosal tears which were repaired with Vicryl 2/0 (Fig 2f). Finally, the whole cavity was inspected for haemostasis and rectal integrity was verified by per rectal examination. The plastic surgeon did bilateral gluteal flaps to fill the dead space and provide support to posterior rectal wall. Wound was closed over two suction drains (Fig 2g). Post- operative neurology was same as pre-operative but on day 14, wound dehiscence occurred at same pre-op non healing area with active discharge. Multiple debridements, culture based (anti-pseudomonal) antibiotics and a total of four vacuum assisted (VAC) sessions followed by a split skin graft finally achieved a complete skin closure over next 8 weeks (Fig 2h).

Histopathology confirmed surgical margins to be marginal. Post-operative imaging both CT and MRI showed complete resection (Fig 3). No further radiation was done as chordoma is radio resistant while proton beam is not available in Pakistan. The patient is 15 months out of his final surgery, can walk unassisted, is able to control his bowel function with laxatives and diet and can maintain continence most of the time for bladder.

Discussion

Primary sacral tumours are relatively rare, and of these chordoma is the most common. Chordoma is slow growing and locally invasive and is usually diagnosed late because of mild and vague symptoms in the early stage. Surgery remains the mainstay of treatment, as these tumours are both radio and chemo resistant. The surgical approach depends on the level involved. For lesions involving S3 and below, posterior alone is sufficient while those located higher than S3 require both anterior and posterior approach. The existing nomenclature for tumour margins in sacrectomy and the level of sacral amputation is depicted in Table 1. The functional outcome is correlated with the extent of roots sacrificed. Generally, L5 and S1 are related to motor, S2 with sphincters, S3 with sexual dysfunction and S4 with perianal anaesthesia [3]. The anococcygeal raphe has no

Table-1: Nomenclature for Sacrectomy with corresponding Recurrence rate and Functional outcome.

Nomenclature for Tumor Margins		Recurrence rate(7)	
		Local	Distant
Contaminated	gross intraoperative violation or positive margins at pathologic analysis	64.7%	75%
Marginal	none of the above-mentioned factors occurred	44.4%	40.0%
Wide	margin of unviolated healthy tissue around the tumour	25.6%	16.4%

Nomenclature for Sacral Amputation (based on lowest nerve root preserved)		Residual Function (3)		
		Ambulation	Bowel	Bladder
High	one S1 preserved	56.2%	3.6%*	2.3%*
Middle	one S2 preserved	100%	12.5%	25%
Low	one S3 preserved	100%	70%	72.7%
Total	Both S1 sacrificed	20.5%†	0%	0%

,2†Indirectly calculated, *Mild function not normal.

role in continence, rarely causes instability.

Although wide marginal enbloc excision is the key as intra-lesional surgery carries 100% recurrence [4], the exact definition of negative margins in Sacrectomy differs from the usual description employed in musculoskeletal system. This is attributed to the proximity of critical structures like rectum, cauda equina, and iliac vessels, which render anything more than a "marginal" resection. Infact, truly the term "wide" margins cannot be applied in enbloc sacrectomy procedures [5]. Regarding biomechanical stability following sacral tumour resection, the sacroiliac stability is good enough as far as 50% or more of the sacroiliac joint is kept intact. However, 30% reduction in load to failure as compared to controls was observed when resection was undertaken between first and second sacral vertebral body [6].

Conclusion

To the best of our knowledge, no text detailing surgical steps of en-bloc sacrectomy is found in indexed Pakistani literature. Moreover, few cases of intralesional biopsies were encountered in our outpatient clinic. We therefore, narrate our experience thus reiterating the importance of technique being widely known, among neuro/ortho/spine surgeons to avoid the non- standard practice of intra-lesional biopsies on such high-grade lesions.

Consent: Consent for publishing the case was obtained from the patient prospectively.

Disclaimer: None.

Conflict of Interest: None.

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Authors' Contributions
SA: Data conception, acquisition, analysis, drafting, revision, final approval.
AI, MN, MRS: Drafting, revision.
AQ: Drafting, revision, final approval.
SS: Data conception, acquisition, analysis, final approval.