



Sacral chordoma: clinical experience of a series of 11 patients over 18 years

Jason Beng Teck Lim¹ · Henry Soeharno¹ · Mann Hong Tan¹

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Abstract

Sacral chordoma are rare low-to-intermediate grade malignant tumours that occur most commonly within the sacrum. Sacrectomy with wide resection margins seems to offer the best long-term prognosis. This study aims to review the management of sacral chordomas including the duration of symptoms, features, treatment, complications and local recurrence rate following surgery at a tertiary centre. We retrospectively reviewed 11 patients treated at our institution between years 1999 and 2015. Patient data included age, sex, history, radiographs, surgical details, onset of recurrence, subsequent treatment, disease-free survival and overall survival were analyzed. Nine patients underwent surgical management with 1 through a sacral approach and eight patients through a combined abdominosacral approach. Despite wide resection in our series, sacral chordoma poses a major problem with approximately 60% of patients having local recurrence in their follow-up.

Keywords Sacral chordoma · Management · Recurrence

Introduction

Chordoma is a rare (1% of malignant bone tumours), insidious and slow-growing malignant tumour, which is thought to arise from the remnants of the notochord [1]. Majority of chordoma 40–50% arise in the sacral region, 30–35% arises from the clivus and 15% occur elsewhere on the axial skeleton [2–5]. It occurs mainly in the fifth to seventh decade of life with a higher incidence in male than in female patients (Ratio of 2:1) [6]. Chordomas are slow-growing tumours; however, they are locally aggressive and often infiltrate into the surrounding tissues and organs [7, 8]. Sacral chordoma often reach a large size and cause late-onset compressive

neurological symptoms when diagnosed [9]. Patients often present with advanced disease due to their vague and indolent symptoms produced by these slow-growing tumours of the sacrum.

Chordomas have a poor sensitivity to radiotherapy and chemotherapy; thus they are mainly treated surgically [2, 10–12]. Complete surgical excision during the initial surgery offers the only therapeutic modality to offer a cure [3]. With widespread infiltration of the chordoma in the sacrococcygeal region, it is often difficult to excise completely; thus, intralesional excisions are often been performed. However, intralesional excisions are associated with high rate of local recurrence and poor prognosis [13, 14]. Sacrectomy with wide resection margins seems to offer the best long-term prognosis. The suitability for patients to undergo en bloc sacral resections with wide surgical margins for complete removal of the chordoma depends on the location of the tumour and surrounding tissues involved [7]. Often there may be extensive nerve root excision leading to high rates of complications such as sexual dysfunction, lower extremity muscle weakness, bowel incontinence and urinary incontinence [7, 15]. The more cephalad levels of resection (cephalad to S2) were associated with poorer urinary and bowel control, especially if bilateral S2 nerve roots are not retained after surgical resection. The higher prevalence of wound

This study was performed at Singapore General Hospital, Singapore, Republic of Singapore.

✉ Jason Beng Teck Lim
jason.lim@mohh.com.sg

Henry Soeharno
henry.soeharno@singhealth.com.sg

Mann Hong Tan
tan.mann.hong@singhealth.com.sg

¹ Department of Orthopaedic Surgery, Singapore General Hospital, Outram Road, Singapore 169608, Republic of Singapore

breakdown occurred before musculocutaneous flap became part of a routine surgical protocol [4].

From various studies [3, 4, 7, 15], the estimated mean 5-year, 10-year and 15-year survival rate range is approximately 80, 60 and 50%, respectively. Local recurrence was associated significantly with an increased risk of metastasis and tumour-related death [3–5, 7, 14–17].

In our study, we aim to review the management of sacral chordomas including the duration of symptoms, features, surgical treatments including flaps performed, complications and local recurrence rate following surgery at a tertiary centre to contribute to the literature for these complex tumours.

Patients and methods

Institutional review board approval was obtained for this study (CIRB number: 2016/2377). We retrospectively reviewed 11 patients treated at our institution between years 1999 to 2015 with up to 15 years (Range 2–15 years) of follow-up. A fellowship trained musculoskeletal tumour senior surgeon at our institution operated on all of the patients. There were six male patients and five female patients. Nine patients underwent surgical resection and two patients were not treated as they declined surgical management. Patient clinical details were analysed which include age, sex, history, radiographs, surgical details, onset of recurrence, subsequent treatment, disease-free survival and overall survival. All of the patients had plain radiographs, radio-isotope bone scan, computed tomographic scan, magnetic resonance imaging. Computed tomographic (CT) scan was obtained to ensure that there were no distant metastases.

Histological confirmation was obtained in all patients. Open biopsy was utilised when the sacral chordoma was palpable and very superficial, or have ulcerated through the skin, which is accessible posteriorly via a posterior midline incision, whereas CT-guided biopsies were performed for more deeply embedded tumours to avoid a major dissection prior to surgical resection. All the biopsy tracts should be incorporated during the primary surgical resection.

Pre-operatively, patients who have involvement of levels cephalad to or including at least a portion of S2 will be planned for a combined sequential anterior and posterior approach with musculocutaneous flap reconstruction for the large, deep soft tissue defect. The location of tumour was the most proximal vertebral level involvement.

For a patient with S3 to coccyx resection, the patient underwent a posterior approach for resection. For surgical margins, wide margin was defined as a cuff of normal tissue around the tumour. The resection margins were considered adequate if it was wide resection based on the intra-operative frozen section as well as post-operative tissue histological assessments. All patients are planned for flap reconstruction

to reduce incidence of wound breakdown and seroma formation for the defect post-surgical resection. Spinopelvic reconstruction will be required for patients who undergo resection cephalad to S1.

Pre-operative angioembolisation will be performed for patients with the presence of large feeding vessels to the sacral chordoma on CT scan to reduce blood loss intra-operatively and reduce the need for ligation of internal iliac vessels. Only the feeding vessels are embolised, and embolisation of the tumour feeding vessels was done under digital subtraction angiography. Post-operatively, patients with marginal resection or histological findings of dedifferentiated chordomas will be planned for adjuvant radiotherapy.

The clinical data of the 11 patients are shown in Table 1. In our study, 73% of patients presented after the fifth decade of life with the mean age of diagnosis at 52.7 years (Range 28–70 years). The mean duration of symptoms is 9.7 months (range 1 month to 2 years). Ten patients had symptoms of lower back pain, seven patients had urinary retention with feeling of incomplete bowel evacuation, one patient had cutaneous sensory loss over foot and one patient presented with symptoms of intestinal obstruction. All of the patients had histological confirmation of sacral chordoma; five patients had open biopsy and six patients had CT-guided trucut biopsy. Two patients had features of dedifferentiation on final histology following surgical resection. The typical features of a sacral chordoma on plain radiographs and magnetic resonance imaging are shown in Figs. 1 and 2.

There are nine patients who underwent surgical management with one through a sacral approach and eight patients through a combined abdominosacral approach. Four patients had adjuvant angioembolisation pre-operatively within a week prior to the resection surgery. The levels of resection at index surgery include eight patients with S2 resection and one patient having S3 resection. There were eight patients who had wide resection and one patient with marginal resection margins. After wide resection of the sacral chordoma, five of our patients had gluteal rotation flap, three patients had vertical rectus abdominis myocutaneous (VRAM) flap and one patient had VRAM flap with free latissimus dorsi myocutaneous flap reconstruction for the surgical defect.

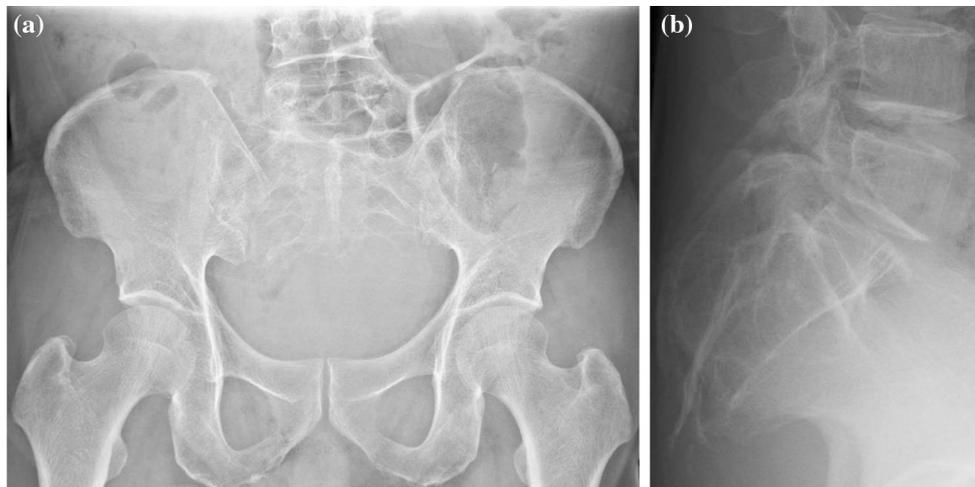
Results

Only 1 (9%) patient in the study group had evidence of metastasis at the time of presentation; this patient had lung metastases detected on CT of the thorax. The other ten patients did not have any distant metastasis at the time of diagnosis and throughout the rest of the follow-up period.

Post-operative complications were encountered in 3 out of the 9 (33%) patients, who had undergone index surgical resection. Two patients developed wound dehiscence that

Table 1 Clinical data for patients with sacral chordoma

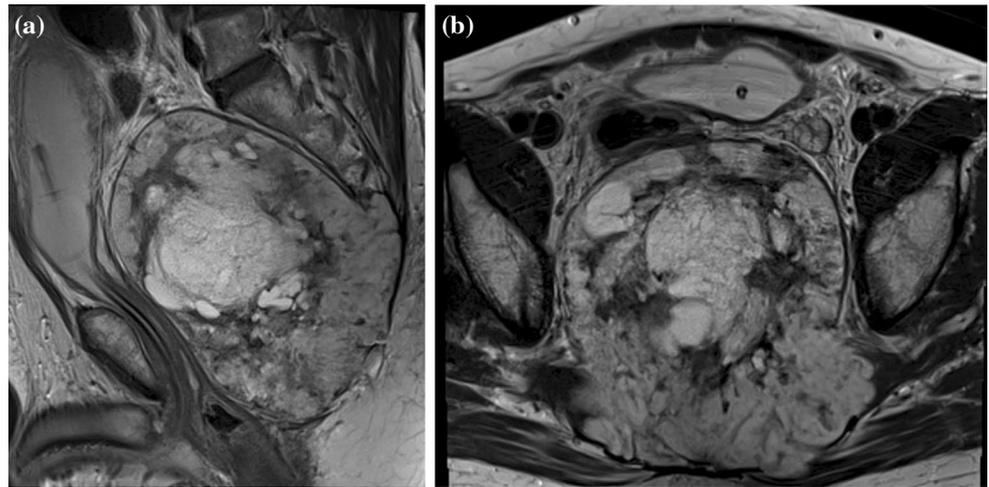
Patient number	Gender	Age of diagnosis (years)	Location	Operative procedure	Histology	Time to first local recurrence (years)
1	Male	58	S3 to coccyx	Wide resection via combined anterior/posterior approach with gluteal rotational flap reconstruction	Conventional Chordoma	Nil
2	Male	70	Left sacral alar	Declined operation	Conventional Chordoma	N.A.
3	Female	43	S1/S2	Declined operation	Conventional Chordoma	N.A.
4	Male	53	S2 to coccyx	Wide resection via posterior approach with gluteal rotational flap reconstruction	Conventional Chordoma	4
5	Male	67	S2 to coccyx	Wide resection via combined anterior/posterior approach with VRAM flap reconstruction	Dedifferentiated Chordoma	Nil
6	Female	54	S2 to coccyx	Wide resection via combined anterior/posterior approach with gluteal rotational flap reconstruction	Conventional Chordoma	2
7	Female	52	S2 to coccyx	Wide resection via combined anterior/posterior approach with gluteal rotational flap reconstruction	Dedifferentiated Chordoma	Nil
8	Female	28	S1/S2	Wide resection via combined anterior/posterior approach with VRAM flap reconstruction	Conventional Chordoma	12
9	Male	53	S3 to coccyx	Wide resection via combined anterior/posterior approach with VRAM and LD myocutaneous flap reconstruction	Conventional Chordoma	Nil
10	Male	58	S2 to coccyx	Wide resection via combined anterior/posterior approach with VRAM flap reconstruction	Conventional Chordoma	1.5
11	Female	43	S2 to coccyx	Wide resection via combined anterior/posterior approach with VRAM flap reconstruction	Conventional Chordoma	3

**Fig. 1** a, b X-ray pelvis (AP) and sacrum (Lateral) showing lytic lesion over sacrococcygeal bone

was managed with wound debridement within 3 months post-operatively and one patient developed intestinal

obstruction, requiring laparotomy and adhesiolysis at 2 years post-operatively.

Fig. 2 a, b MRI pelvis (T2 W) showing large sacral tumour with central necrosis compressing on anterior pelvic viscera



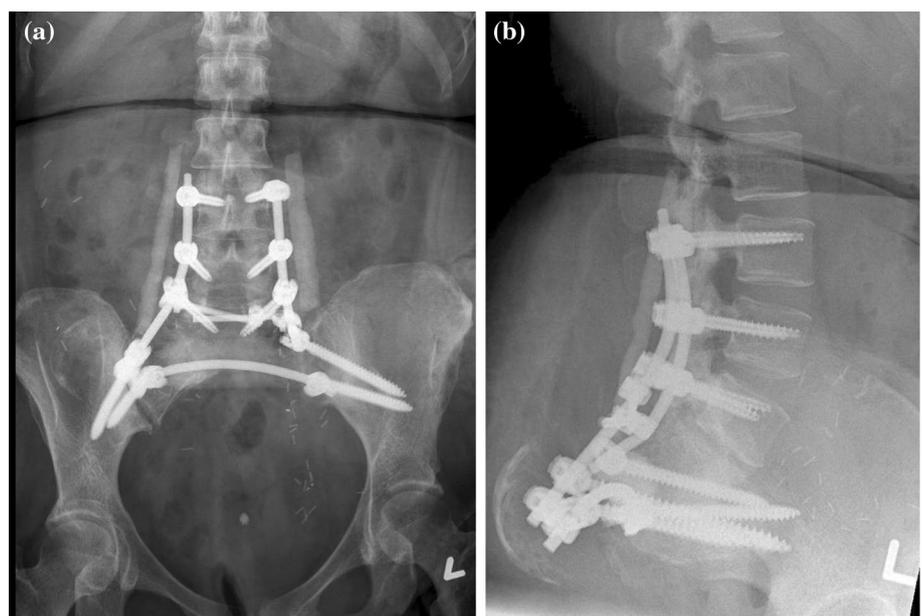
The rate of local recurrence in our study is 45%, occurring in 5 out of the 11 patients. The mean duration at which recurrence occurred was 4.5 years (range 1.5–12 years) from the time of the first surgical resection. Of the five patients with local recurrence, 2 declined surgical intervention. The other three patients underwent repeated wide resection of the recurrence; 2 of them underwent a combined anterior/posterior wide resection of the recurrence, while the last patient required en bloc resection of the L5/S1 due to the large recurrence and had L3 to ilium posterior instrumentation (Fig. 3) with rotational gluteal flap of the sacral defect. There was no significant complication associated with the spinopelvic reconstruction, and the patient was able to ambulate and weight-bear post-operatively. Of the three patients who underwent surgical resection for local recurrence, 2 had recurrence within 2 years of the second surgery.

At the end of the follow-up period, 7 out of the 11 (63%) patients remained alive. The two patients who had declined surgical management demised at 2 and 12 years after diagnosis. Two of the nine patients who underwent an index surgical resection demised at 2 and 7 years post-operatively. The mean disease-free interval is 5.4 years from the time of surgery to the end of the follow-up period in December 2015 (range 2–15 years follow-up).

Discussion

Sacral chordoma is the most common primary sacral tumour, followed by the giant cell tumour. It can be difficult to differentiate between the 2 lesions as they share several similar characteristics; however accurate diagnosis is important so

Fig. 3 a, b X-rays lumbar Spine (AP/Lateral) showing L3 to ilium posterior instrumentation and fusion with femoral allograft, pedicle and iliac screws post-en bloc wide resection of L5/S1 sacral chordoma



as to determine best treatment. A scoring system has been reported by Tsuji et al. [18] based on clinical characteristics and radiological findings, which can be very helpful to differentiate between the 2 commonest sacral tumours. The six items included in the scoring system [18] include patient's age, site of occurrence, tumour expansion pattern, T2-weighted images, septal structure, and incomplete bone shells, which were significantly different between giant cell tumour and sacral chordoma patients. All our patients underwent pre-operative CT and MRI for pre-operative planning and staging. In addition, it is important to obtain a tissue sample for biopsy in all patients presenting with a sacral tumour, thus all of our patients had pre-operative biopsies to obtain histological confirmation of a sacral chordoma before surgical resection was planned.

Surgical excision with wide margins is necessary for treatment of sacral chordoma due to its poor sensitivity to radiotherapy and chemotherapy [4, 16]. The extensive nature of the resection makes this a technically challenging procedure, and there are many pre-operative and intra-operative considerations that should be taken into account to minimise post-operative complications and recurrences.

For proximal sacral resections above the S3 vertebra, the resection should be performed by a combined anterior and posterior approach. The anterior approach would allow for identification of pelvic structures, mobilisation and ligations of branches internal iliac arteries and exposure of the sacrum proximal to the tumour to for osteotomy to be performed. The posterior approach when utilised for proximal sacral resections has risk of possible haemorrhage and pelvic viscera injury during the tumour resection.

We recommend routine flap reconstruction for all patients undergoing wide resection of sacral chordoma, with appropriate pre-operative planning in conjunction with plastic surgeons. This is because studies have shown lower rates of wound breakdown when reconstruction with flaps is routinely performed for sacral chordoma [15, 19, 20]. Indeed, all nine of our patients undergoing wide resection were assessed by plastic surgeons pre-operatively and had flap reconstruction immediately following wide resection. Two out of the nine patients in our series experienced a post-operative complication related to flap reconstruction. Both patients only had superficial wound dehiscence, which was successfully managed with wound debridement. In addition, all patients were operated through a combined abdominosacral approach, which has been reported in the literature to offer the safest and maximal exposure for flap reconstruction [21].

Wide resection margins are a very important prognostic factor that has been shown to correlate with the incidence of local recurrences and overall survival. Nonetheless, local recurrence is common even after wide resection. In our series, the rate of local recurrence was 45%

despite wide resections in eight out of the nine patients who underwent surgery. Some reports have suggested that local recurrence after a wide resection may be related to infiltration of adjacent musculature or microscopic satellite lesions which would not have been detectable by imaging [22]. We recommend that after successful en bloc resection of the tumour, intra-operative frozen sections of osteotomy sites, sacroiliac joints and musculature adjacent to the sacrum such as glutei and piriformis muscles to be performed to evaluate for infiltration and further resection can be performed.

Aggressive variants of sacral chordomas have been reported which have rapid clinical deterioration and metastasis, with a high risk of recurrence within a short period of time, usually within 2 years, even after wide resection with clear margins. Some of these aggressive tumours have been found to be dedifferentiated chordomas, such as dedifferentiation to the fibrosarcoma, osteosarcoma and malignant fibrous histiocytoma [23, 24]. Other aggressive variants of sacral chordomas include those with mitotic figures and/or Ki-67 labelling indices more than 5%, tumours with microsatellite instability and those with chromosomal abnormalities [3, 25, 26]. Klinger et al. [25] showed that a patient with a loss of heterozygosity in 9p and 18q developed widespread metastasis in a short period of time, while Horbinski et al. [26] showed that a chordoma with 9p21 homozygous deletion also demonstrated an aggressive clinical course [25, 26]. In our study, two patients had dedifferentiated chordomas. Both patients did not have any distal metastasis throughout the study period. One had adequate surgical margins and achieved a disease-free interval of 15 years, whereas the other had a marginal resection and developed local recurrence within 1.5 years. The second patient's recurrence may be related to the marginal resection rather than the dedifferentiated nature of the tumour. Nonetheless, it is important for clinicians to recognise that while most sacral chordomas are slow-growing, some have the potential to cause rapid deterioration. Hence, clinical vigilance and close follow-up is recommended.

As reported in the literature, we recommend that surgical resection is warranted in local recurrence and a complete resection of local recurrence offers the best chance of control of disease [27–34]. Local recurrence and metastasis rates in other studies are shown in Table 2. Rarely can patients who develop recurrence be made disease-free which is likely due to the satellite infiltrations of sacral chordoma [22]. In our series, there were two patients who had re-recurrence within 2 years after repeated wide resection. Local recurrence is often detected on surveillance imaging and also based on the recurrence of symptoms experienced by patients. It has been reported that the pre-operative neuropathic pain is significantly related to post-operative neuropathic pain and the recurrence of severe pain with different characteristics after

Table 2 Mean follow-up, local recurrence and metastasis rates of sacral chordomas. Tumour-related deaths are patients who demised with persistent tumour, or death related to metastases of chordoma

Authors	No. of patients	Mean follow-up period	Local recurrence	Metastasis	Tumour-related death
Fuchs et al. [4]	52	7.8 (2.1–23)	23 (44.2%)	16 (30.8%)	19 (36.5%)
Bergh et al. [3]	39	8.1 (0.2–23)	17 (43.6%)	11 (28.2%)	10 (25.6%)
York et al. [8]	27	3.6 (0.3–34)	18 (66.7%)	7 (25.9%)	15 (55.6%)
Hulen et al. [15]	16	5.5 (1.25–14.5)	12 (75%)	6 (37.5%)	6 (37.5%)
Yonemoto et al. [7]	13	6.3 (0.5–13.7)	6 (46.2%)	6 (46.2%)	7 (53.8%)
Ruggieri et al. [29]	56	9.5 (3–28)	24 (45%)	16 (30%)	17 (32%)

the sacral chordoma resection should alert clinicians as it may indicate local recurrence of tumour [35].

Resection of sacral chordoma that involves caudal sacral vertebra or sacroiliac joints (SIJs) will cause significant mechanical compromise, and surgical planning should include the need for potential internal fixation post-resection [36–39]. Tumour resection between first and second sacral vertebra significantly reduced stability in 50% of patients and fatigue fractures may complicate more caudal sacral resections. In the setting of total sacrectomy, the use of double-rod double iliac screw method provided the most rigid fixation for satisfactory mechanical support and ambulation for the spinopelvic dissociation after total sacrectomy [37, 38]. However, the load to failure after caudal resection may often exceed the physiological loads of the patient. In a recent study by Kiatisevi et al. [40], they report a series of 16 patients with good functional outcome after total sacrectomy without spinopelvic reconstruction, especially for patients who underwent Type I iliosacral resections (medial to or through or lateral but close to the sacroiliac joint). Without spinopelvic reconstruction, most patients in their series [40] were able to walk with good Musculoskeletal Tumor Society scores (MSTS). Total sacrectomy without spinopelvic reconstruction should be considered as a useful alternative to reconstructive surgery in patients who undergo Type I iliosacral resection on one or both sides to avoid the complications associated with spinopelvic reconstruction. Similarly, in our series, a majority of our patients did not require spinopelvic reconstruction even for caudal resection for resection between S1 and S2. There was only one patient who required spinopelvic reconstruction post-resection due to local recurrence requiring en bloc resection of L5/S1. We recommend spinopelvic reconstruction only if sacroiliac joint is resected and for proximal resection with L5 involvement.

Sacral chordomas are low- to intermediate-grade malignant lesions that present late in the disease process due to their vague and indolent symptoms. To date, surgery with wide resection margins offers the best long-term prognosis for these patients. Optimal radiotherapeutic regimes with long-term survival have not been developed, and chordomas remain resistant to conventional chemotherapeutic

agents. The effective management of patients with sacral chordoma includes early diagnosis with wide resection and close follow-up due to high rates of recurrence even after wide resection.

Compliance with ethical standards

Conflict of interest All authors declare that there is no conflict of interest for this study.

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