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Case Report

Recurrent Gluteal Sarcoma Managed with Unilateral Excision of Levator Ani Muscle: Case Report

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ABSTRACT

Soft Tissue Sarcomas (STS) are rare malignant tumors that arise from mesenchyme, 80% arise in soft tissue while 20% in bone, and they comprise 1% adult tumors. Gluteus maximus STS is site with frequently diagnoses of high- and low-grade STS. Low-grade STS respond well to surgery alone while high-grade STS require preoperative chemoradiation therapy, followed by surgery, and then postoperative chemotherapy. Work-up includes: a core needle biopsy for histopathological diagnosis, MRI for imaging of local disease and Contrast enhanced CT scan for pulmonary metastasis. Recurrence is viewed as a sign of poor local treatment and a risk for distant metastasis. Reduction of local recurrence does not lead to improved survival, but lack of disease progression with pulmonary metastasis does. In our patient, laparoscopy allowed total mesorectal excision dissection and sparing of rectum, as there was no metastatic spread of tumor to the rectum. Despite excision of right levator ani muscle, our patient maintained her continence, as shown by Fucini *et al.* [1] that continence would be maintained despite dissection and separation of levator ani muscle from the anal complex unit (external and internal anal sphincter) followed by unilateral excision of levator ani muscle, while achieving good oncologic and anal function outcome. We present our management of a 55-year-old lady with recurrent gluteal STS with extension into the ischioanal fossa managed at Tata Memorial Hospital, in Parel, Mumbai, India, in the Department of Colorectal Surgery.

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Introduction

Soft Tissue Sarcomas (STS) are rare malignant tumors that arise from mesenchyme. 80% of STS arise in soft tissue while 20% in bone, and they comprise 1% of adult tumors [2-5]. Though the cause of STS is unknown, they have heterogeneity with consideration to age of onset, age distribution, anatomic location, disease progression and outcome [2-4]. The Gluteus maximus STS is site with frequent diagnoses of high- and low-grade STS [5, 6]. Low-grade STS respond well to surgery alone while high-grade STS require preoperative chemoradiation, followed by surgery, and postoperative chemotherapy [2, 3]. High-grade STS have 50% potential to metastasize and 5-year overall survival [3].

Clinical features are varied due to heterogeneity. Presentation of a gluteal STS is usually late resulting in delayed diagnosis and treatment, due to large size of gluteus maximus muscle and its ability to accommodate large tumors, hence, gluteus maximus muscle has been referred to as silent area for large tumors [2]. High suspicion index for malignancy should be applied to STS with increasing size, size above 5cm, painful and location that is deep to deep fascia [3]. Work-up includes: core needle biopsy for histopathological diagnosis, MRI imaging of local disease and Contrast enhanced CT scan for pulmonary metastasis [2, 3]. We present our management of a 55-year-old lady with a recurrent right gluteal STS that had extension into the right ischioanal fossa, managed at Tata Memorial Hospital, in Parel, Mumbai, India, in the Department of Colorectal Surgery.

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Case Report

A 55-year-old lady presented with pain and swelling in the right gluteal region of 1.5 years duration. She was evaluated for the same with a contrast enhanced computerized tomography scan (CECT) that showed a 15cm mass in the right gluteus muscle with extension into ischiorectal fossa. Fine needle aspirate showed a spindle cell tumor following which she underwent a piece meal, incomplete, margin positive excision. She then presented to our hospital where pathology of the excised tumor was reviewed to be low grade myxofibroid sarcoma. Magnetic resonance imaging (MRI) of the pelvis revealed residual tumor in the right ischiorectal fossa abutting the right levator ani muscle (Figure 1).

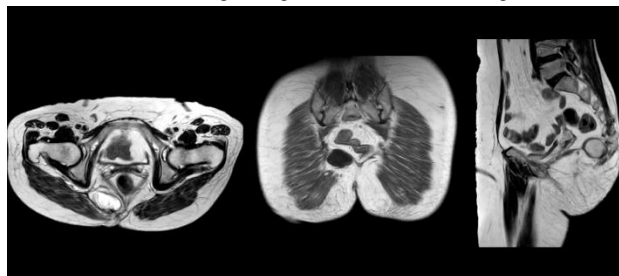


Figure 1: MRI images showing T2 hyperintense, T1 hypointense tumor abutting in the ischiorectal fossa abutting the right levator muscle.



Figure 2: Skin incision around previous suture line and drain site.

A CT scan of the thorax was negative for metastasis. She was planned for a repeat wide excision of the residual mass. To protect the anorectum from injury during the excision, a laparoscopic rectal mobilization was performed with placement of a pack posterior to the rectum and over the tumor as visualized from the abdomen. Patient was turned prone and skin incision was marked to excise the previous suture line and eccentric drain site en-bloc (Figure 2). Skin and subcutaneous flaps were elevated and the gluteus maximus was widely divided after protecting the sciatic nerve in the retro-gluteal bursa. In the midline, S5-Coccyx junction was divided followed by division of the anococcygeal ligament. The pelvis was entered to retrieve the pack placed during the laparoscopic rectal mobilization (Figure 3). The sciatic nerve was followed proximally till its exit from the greater sciatic notch after which the inferior gluteal and internal pudendal vessels were divided (Figure 4). The levator was then divided taking adequate margins around the tumor. Laterally ischial spine and ischial tuberosities formed the limits of excision and the sacrotuberous ligament was divided (Figure 5).

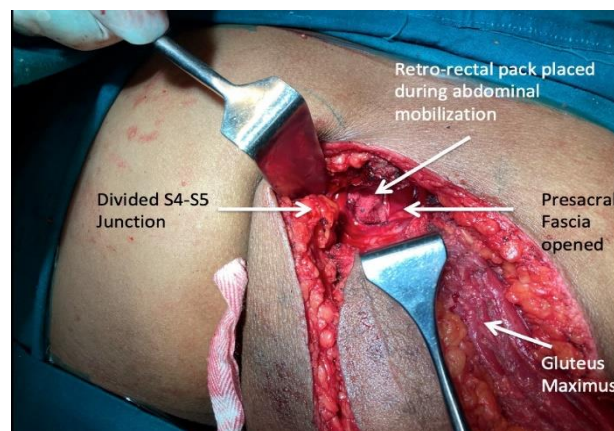


Figure 3: Retrieval of abdominal retro-rectal pack after coccyx and anococcygeal ligament division.

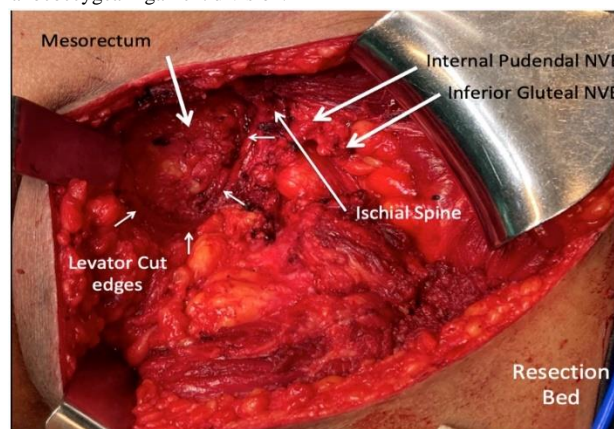


Figure 4: Resection bed.

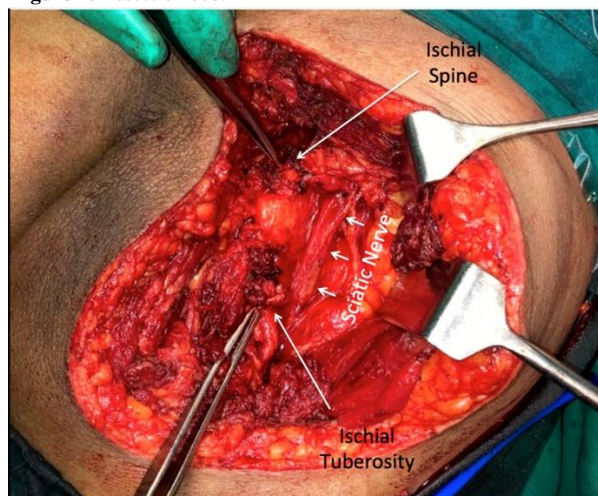


Figure 5: Exposed sciatic nerve till its exit from the greater sciatic notch.

After the delivery of specimen, the remaining gluteus muscle was mobilized and sutured to cover the exposed ano-rectum followed by subcutaneous and skin closures over drains (Figure 6). She had an uneventful post-operative recovery and was discharged on post-operative day 3. Final histology revealed Myxoid liposarcoma with negative margins (R0 resection). Patient had satisfactory fecal continence and is planned for adjuvant radiation in view of its large size (15cm), deep location and prior unplanned, piece-meal excision putting her at high risk for local relapse.



Figure 6: Skin closure.

Discussion

STS are rare tumors, making up 1% of adult tumors. The diagnosis of STS is often delayed, making them grow to large size with a presentation of discomfort and/or constipation at the time of diagnosis [6, 7]. While tissue diagnosis by core needle biopsy is required, our patient had a recurrent tumor and the diagnosis was confirmed by fine needle aspiration cytology [3]. Even though fine needle aspiration cytology is not recommended as a diagnostic modality, it is useful to confirm recurrence of the tumor [3]. Recurrence is viewed as a sign of poor initial local treatment, and a risk for distant metastasis. However, reduction of local recurrence does not lead to improved survival, lack of disease progression with pulmonary metastasis does [8]. Therefore, a patient with local recurrence and without concurrent distant metastasis has a prognosis same as one with microscopic negative margins (R0 resection) without recurrence [3, 8]. However, there is no consensus on the rate of recurrence for STS [8]. Our patient had recurrence of right gluteal sarcoma extending to the right iliac fossa confirmed on MRI imaging. MRI is the preferred imaging for tumor location, size, and relationship to neighboring organs and in our patient the tumor was in right iliac fossa abutting right levator ani muscle [3, 7]. Contrast enhanced CT scan was negative for distant metastasis.

Therefore, abdominoperineal approach with laparoscopy was used for its benefit of optimal pelvic-floor visualization, dissection, and assessment of levator ani muscle resection, which is not easily done in open abdominal surgery [1, 9, 10]. In our patient, laparoscopy allowed total mesorectal dissection and sparing of rectum, as there was no metastatic spread of tumor to the rectum. Despite excision of right levator ani muscle, our patient maintained her continence, as shown by Fucini *et al.* that continence would be maintained despite unilateral dissection and separation of levator ani muscle from the anal complex unit (external and internal anal sphincters) followed by unilateral excision of levator ani muscle, while achieving good oncologic and anal function outcome [1]. This technique by Fucini *et al.* avoids resection of rectum with tumor free margins that leaves wide pelvic defects requiring reconstruction of pelvic floor and a permanent colostomy [1]. APR is still recommended for tumors that involve both levator ani muscles, those with recurrent tumors that extend to right iliac fossa, and those in whom R0 resection cannot be achieved with unilateral levator ani muscle excision due to

preoperative treatment or distortion of anatomy by tumor recurrence [1, 6].

Therefore, initial surgery to achieve R0 resections with rim of normal margins, must be planned after discussion in MDT and need for radiotherapy, whether pre- or post-operative assessed [3, 8]. Adjuvant radiotherapy is indicated for intermediate and high grade tumors and those with large tumors more than 8cm in size that were marginally excised, or recurrent low-grade tumors [3, 11]. However, adjuvant radiotherapy is avoided in those with low-grade tumor and there is no evidence to support the benefit of adjuvant chemotherapy [3]. Our patient recovery was uneventful and has been planned for adjuvant radiotherapy based on tumor size and recurrent nature of the disease. Postoperative follow-up for our patient will include clinical details of history and physical examination, with primary tumor site evaluation and imaging, and chest imaging every 3-6 months for 2 to 3 years, followed by every 6 months for next 2 years [3, 12].

Conclusion

STS are rare tumors and wide resection remains the preferred standard treatment with chance of cure. Lack of surveillance programs for STS increases incidence of recurrence and disease progression to distant metastasis. Our case has shown that unilateral levator ani muscle resection can be done with acceptable functional and oncological.

Conflicts of Interest

None.

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