

CASE REPORT **OPEN ACCESS**

A Ten-Step Surgical Approach to Radical Excision of a Primary Ischiorectal Fossa Tumor With IGAP V-Y Flap Reconstruction—A Case Report of a Rare Myxofibrosarcoma

Amy Whitmore¹  | Sabina Ioana Nistor² | Muhammad Ather Siddiqi³ | Roman Mykula⁴ | Hooman Soleymani majd^{2,5} 

¹University of Oxford, Oxford, UK | ²Department of Gynaecology Oncology, Churchill Hospital, Oxford University Hospitals National Health Service (NHS) Foundation Trust, Oxford, UK | ³Nuffield Orthopaedic Centre, Oxford University Hospitals Foundation Trust, Oxford, UK | ⁴Plastic Surgery, Oxford University National Health Service (NHS) Foundation Trust, Oxford, UK | ⁵Nuffield Department of Women's and Reproductive Health, University of Oxford, Oxford, UK

Correspondence: Hooman Soleymani majd (hooman.soleymanimajd@msd.ox.ac.uk)

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ABSTRACT

This case report presents a rare case of ischiorectal fossa myxofibrosarcoma, which was successfully excised with a clear margin and full preservation of the patient's pudendal nerve function. We present our surgical technique in 10 steps, describing the surgical excision and reconstructive surgery with the inferior gluteal artery perforator V-Y flap.

1 | Introduction

The ischiorectal fossa (IRF) is an often neglected anatomical space, unlikely to be at the forefront of most surgeons' minds. We believe it should be considered an important area in multiple different surgical disciplines. The IRF is the largest space in the anorectal region [1]. It hosts a variety of vessels and nerves, including the internal pudendal nerve and vessels, inferior rectal nerve, the perineal branch of S4, and perforating cutaneous nerves [2, 3]. This, combined with the proximity to functionally important structures such as the anal canal, levator ani, and the attachment of the external anal sphincter to the coccyx [2], makes it an area of surgical importance that should be considered by any team operating within this region.

Though perhaps most thought of in the context of abscess drainage [4], the IRF can harbor a number of different primary

tumors, arising from any of the tissues in the fossa, as well as growth of metastatic tumors (Table 1) [1]. There are currently no clinical guidelines on the management of primary tumors, malignant or benign, located in the IRF, and although these tumors are very rare [5–7], any surgical team that encounters them should aim toward optimal treatment for patients affected by pathology of this space.

1.1 | Anatomy Considerations

The IRF has a pyramidal shape and it is bordered superomedially by the levator ani muscle, inferomedially by the external anal sphincter, laterally by the obturator internus muscle, inferiorly by the perineal skin, anteriorly by the superficial and deep transverse perineal muscles, and posteriorly by the lower border of the gluteus maximus muscle and the sacrotuberous ligament

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(Figure 1 and Table 2) [1]. The left and right IRF communicate posteriorly via the post-anal deep space, between the levator ani and the anococcygeus ligament [14].

The IRF's blood supply is provided by the rectal and labial branches of the internal pudendal vessels, while the innervation is via the pudendal nerve. This neuro-vascular bundle travels in the pudendal canal, on the lateral side of the IRF, just medial to the obturator internus fibers. The lymphatic drainage of the region is to the internal iliac lymph nodes [18].

1.2 | IRF Tumors

Tumors of the IRF are rare, and evidence in the literature consists predominantly of case reports and case series. The largest published series by Zhu et al. [11] comprises 24 patients, 61% of whom had benign pathology. There were no cases of aggressive angiomyxoma in this series—pathology previously reported as the most common primary tumor of the IRF [18]. The histopathology of IRF tumors is heterogeneous, as benign and malignant neoplasms arise from the different anatomical structures that form the fossa, such as blood vessels, nerves, muscle, and adipose tissue, skin [1]. We provide a list of the most common tumors affecting this area in Table 1.

Of note, the pathology of the IRF differs significantly from that of the pre-sacral space, dominated by mostly benign pathology—congenital or developmental cysts and tumors due to its midline location and the close proximity of several organ systems during embryological development [11].

Patients affected by IRF tumors are predominantly female, in the middle-aged range [11].

Imaging is essential in describing IRF tumors. Computed tomography (CT) and magnetic resonance imaging (MRI) are both useful in describing features such as location, size of the tumor, relationship to surrounding structures, tumor characteristics, and potential diagnosis [1]. Fluoro-deoxy-glucose Positron Emission Tomography (FDG PET) is also useful in the

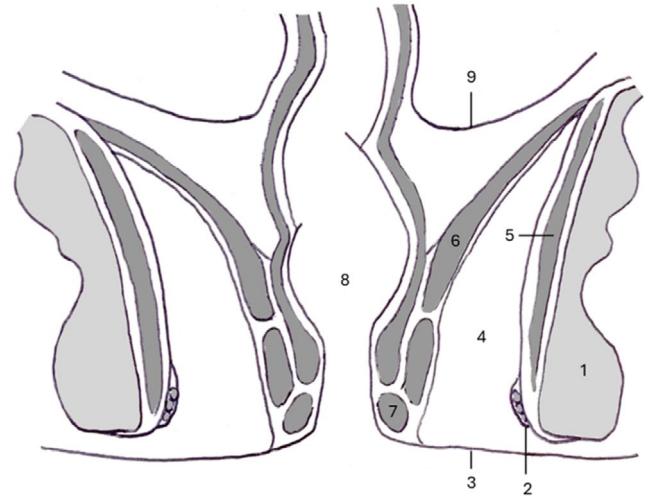


FIGURE 1 | Anatomy of the IRF. 1 Ischial tuberosity. 2 Pudendal canal. 3 Perineal skin. 4 Ischioanal fossa. 5 Obturator internus. 6 Levator ani. 7 External anal sphincter. 8 Rectum. 9 Peritoneum.

TABLE 1 | Benign and malignant neoplasms of the ischioanal fossa, classified by tissue origin [1, 8].

| Ischioanal fossa tumors | | |
|-------------------------|--|--|
| Origin | Benign | Malignant |
| Blood vessels | Aggressive angiomyxoma [9], hemangioma Vascular malformations (Klippel-Trénaunay Syndrome) [10] | Angiosarcoma |
| Nerves | Plexiform neurofibroma (NF1) Schwannoma [5] | Malignant granular cell tumor Malignant peripheral nerve sheath tumor Neuroendocrine tumor [11] |
| Muscle | Solitary fibrous tumors [12] Leiomyoma | Leiomyosarcoma [8] Rhabdomyosarcoma [13] Malignant PEComa Undifferentiated pleomorphic sarcoma |
| Connective tissue | Spindle cell tumor—benign variant [11] | |
| Adipose tissue | Lipoma [6] | Liposarcoma |
| Skin/skin appendages | Hidradenoma papilliferum Proliferating trichilemmal tumor [14] | Merkel cell carcinoma [11] Basaloid carcinoma [11] Bartholin gland carcinoma [15] |
| Other | Inflammatory/infectious processes Other: tailgut cysts, perineal epidermal cyst, dermoid cyst [16], perineal endometriosis [17], and Gardner's duct cysts | Hematogeneous metastases from melanoma, lymphoma, GIST, and chordoma Direct invasion from adjacent pelvic organs, i.e., anorectal or pelvic bone cancer |

TABLE 2 | Anatomical borders of the ischiorectal fossa [1, 18].

| Ischiorectal fossa—anatomical borders | |
|--|---|
| Superior and dorsal | Levator ani muscle |
| Medial lower pole | External anal sphincter |
| Lateral | Obturator internus muscle and obturator fascia |
| Anterior | Superficial and deep transverse perineal muscles |
| Posterior | Lower border of gluteus maximus muscle and the sacrotuberous ligament |
| Inferior | Skin of the perineum |

differentiation between benign and malignant tumors and in assessing the extent of disease, that is, lymph node involvement, metastatic lesions.

Tumors of the IRF are challenging due to their location, which is relatively difficult to access. A posterior, trans-gluteal approach has been described, with the patient in the prone position [19] and is particularly useful when involvement of the sacrum-coccyx is suspected and excision may be necessary. In some cases, a combined anterior abdomino-perineal approach is necessary, particularly when the levator ani muscle is involved [6]. Resection of the rectum may also be required, and patients should be prepared for this pre-operatively. Tumors located completely below the levator ani muscle benefit from a perineal approach, with the patient in the lithotomy position, as the pelvic floor pushes the mass towards the skin [11]. Perineal reconstruction may be required.

The main surgical principle is excision of the mass in its entirety, with a safety margin of tissue, leaving no visible tumor behind (R0). In the series published by Zhu et al. [11], the recurrence rate was 44% in the malignant subgroup, and 75% of recurrences occurred in cases where R0 was not achieved.

Here we present a case report of a female patient with a myxofibrosarcoma of the IRF, with a 10-step multidisciplinary surgical approach to management of this tumor, incorporating both excision of the mass and flap reconstruction of the fossa in a single operation using a perineal approach.

2 | Case History/Examination

This case report focuses on a 48-year-old female patient, referred to the Oxford University Hospitals Trust Gynecology Oncology service from a district general hospital with a 3-month history of an uncomfortable, growing swelling in the left buttock. She was a para 2 with a past medical history of mild asthma using a Salbutamol inhaler when required. Her BMI was in the normal range; she has never smoked and only occasionally drinks alcohol. She had no pre-operative functional restrictions. She had regular, normal cervical smears.

3 | Investigations

An ultrasound scan identified a 9 cm heterogeneous mass with increased internal vascularity. An MRI described an ovoid tumor in the IRF measuring 6.8 × 7.8 × 8.4 cm with features suspicious of a sarcoma (Figure 2). The patient was referred to the Oxford Sarcoma MDT.

An ultrasound-guided biopsy was carried out, and morphological features were in keeping with a Grade 2 myxofibrosarcoma. Immunohistochemistry was negative for AE1/3, SOX10, desmin, MSA, and MDM2, with a non-specific cytoplasmic reaction for STS6. The Ki-67 proliferation index was 10%, which, in the context of soft tissue tumors, demonstrates an intermediate likelihood of high-grade, aneuploid, necrotic tumors [20]. A whole body PET scan (Figure 3) showed no nodal involvement or metastatic disease (pre-treatment TMN staging: T2N0M0), making this patient a suitable candidate for radical surgical management.

The patient received 6 weeks of pre-operative radiotherapy to the left buttock. She developed mild skin erythema in the perineal region following radiotherapy, and surgery was postponed by 2 weeks in order to allow full skin healing. A post-radiotherapy MRI found that there had been a mild decrease in the size of the tumor to 6.4 × 7.4 × 8.4 cm, with a few new foci of hemorrhage within the tumor, displacing the rectum medially, abutting the left obturator internus muscle anteriorly, and the anterior margin of the medial left gluteus maximus muscle posteriorly.

4 | Treatment—The Surgical Approach

The operation consisted of a perineal approach to a radical resection of the left IRF tumor with wide margins. This was followed by perineal reconstruction using an inferior gluteal artery perforator (IGAP) flap with V-Y advancement to close the space left by wide local excision in an attempt to optimize residual function of the perineal region, done in the same operation, to allow for a more comfortable and convenient healing process for the patient [5]. The excision of this large tumor and the local margins did leave a significant residual space, with deviation of the perineum evident after removal of the tumor. An IGAP flap was chosen as the flap of choice for primary closure in this procedure, to fill the residual space and aim to reduce complications from the phenomena known as “empty pelvis syndrome”, which can result in bowel herniation and even bowel obstruction in some cases [21, 22]. Flap reconstruction promotes healing by advancing well-vascularized tissue into a previously irradiated area. This case presents a successful multidisciplinary approach to these surgical techniques, with no complications for the patient to date.

4.1 | Step 1—Examination Under Anesthesia and Planning of Surgical Incision

The patient was placed in lithotomy position, with legs in stirrups and FlowTrons on.

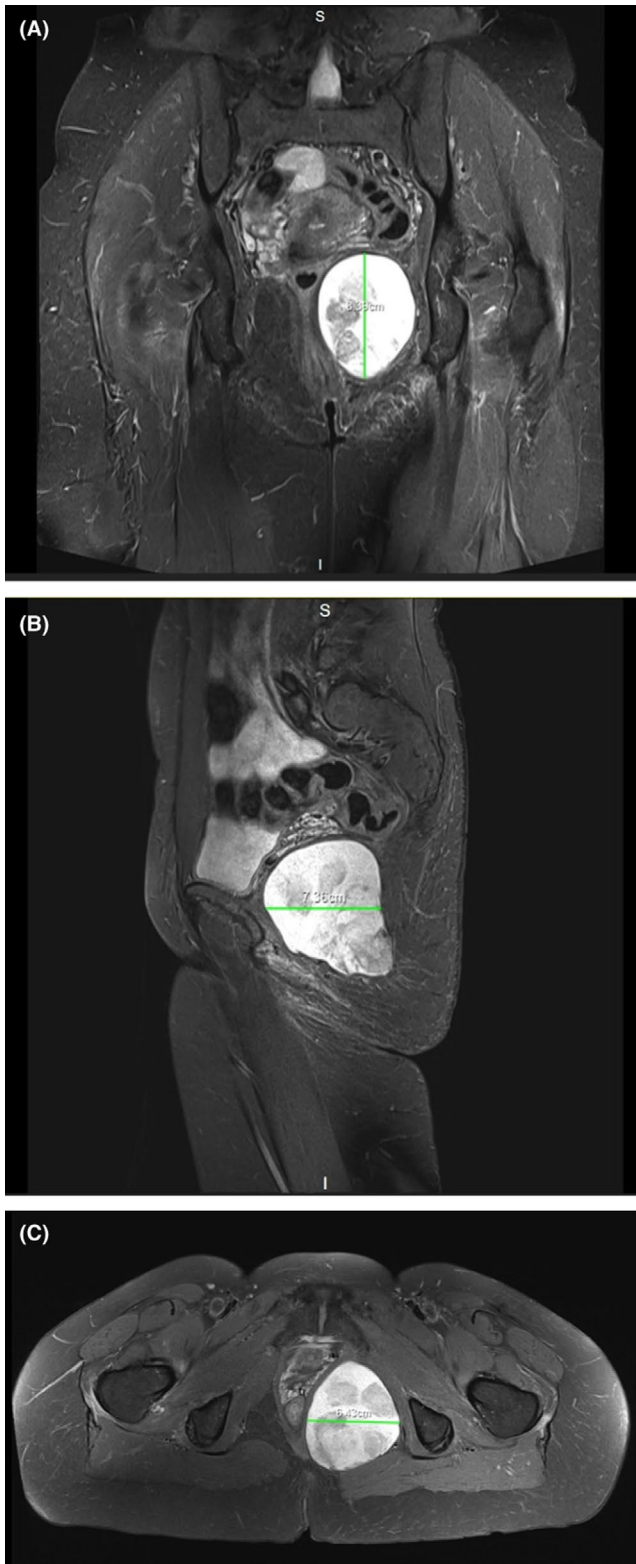


FIGURE 2 | T2-weighted images of the patient's pelvis demonstrating the IRF tumor in situ, sized $8.39 \times 7.36 \times 6.43$ cm; (A), coronal view; (B), sagittal view; (C), axial view.

Examination under anesthesia per vaginum and per rectum found a large left IRF tumor bulging into the left vaginal wall, deeply seated in the pararectal space, abutting the sciatic notch,



FIGURE 3 | PET CT images demonstrating the tumor in situ, showing enhanced metabolic activity localized to the tumor; (A), coronal view; (B), axial view.

and medial to the ischial tuberosity. The aim of this examination was to determine the size and location of the mass in order to guide incisional planning. It was decided that the easiest way to access the tumor was via a transperineal approach.

4.2 | Step 2—Incision of the Skin and Soft Tissues (Figure 4)

The patient was cleaned, draped, and catheterized. A vertical, antero-posterior perineal incision was made with a cold blade, 4 cm left of the vaginal introitus, planned jointly.

The superficial tissue was dissected using monopolar diathermy and Ligasure Exact Dissector until the tumor was reached.

4.3 | Step 3—Assessment of the Depth of the Tumor After Incision

Following dissection of superficial soft tissues, the tumor was revealed and its location more accurately assessed.

4.4 | Step 4—Excision of Tumor With Careful Conservation of Tissue Margins (Figure 5)

Once the tumor had been identified, the tissue surrounding the tumor was dissected, with care taken to not breach the tumor and to remove it with a margin of healthy tissue, but also to not compromise any important surrounding structures. Most importantly, the pudendal nerve, perineal body, external and internal anal sphincters were kept intact, to preserve sexual function and continence [3]. The dissection plane was inferior to the obturator membrane and foramen. A rectal probe was inserted for dissection of the medial plane to ensure identification of the rectal wall to minimize damage to this structure.



FIGURE 4 | The initial incision of the skin, with a cold blade. The incision is vertical and was cut from anterior to posterior, 4 cm left of the vaginal introitus. The position of this incision was jointly planned between all teams involved.

4.5 | Step 5—Devascularization Superficial and Deep (Figures 5, 6)

The tumor was devascularized in a controlled manner using LigaSure Exact.

4.6 | Step 6—Complete Mobilization of the Tumor With Clear Margins (Figure 7)

The encapsulated tumor, measuring 8.5×7.5×6.5 cm, was removed intact and with adequate margins (Figure 8). The patient was re-draped and cleaned. The procedure was handed over to the Plastic Surgery team.

4.7 | Step 7—IGAP Flap Planning and Incision

The IGAP flap was designed on perforator blood vessels within the gluteal fold donor site.

The perforator vessels were located using Doppler to ensure adequate blood supply of the flap.

4.8 | Step 8—IGAP Flap V-Y Mobilization

After the initial incision, the IGAP flap was raised subfascially down to the gluteus maximus muscle using monopolar diathermy. Perforators were isolated and dissected further to allow the flap to be fully mobilized (Figures 9, 10). Once the flap was fully mobilized, 3–6 cm of the anterior edge was de-epithelialized (anteriorly to posteriorly) to allow for adequate filling of the space left behind by the surgical excision of the tumor (Figure 11).

4.9 | Step 9—Securing the Position of the Flap and Drains

The flap was elevated and advanced 7 cm medially, and the de-epithelialized portion of the flap was inserted into the IRF (Figure 12) to fill the empty space (Figure 13). The flap was

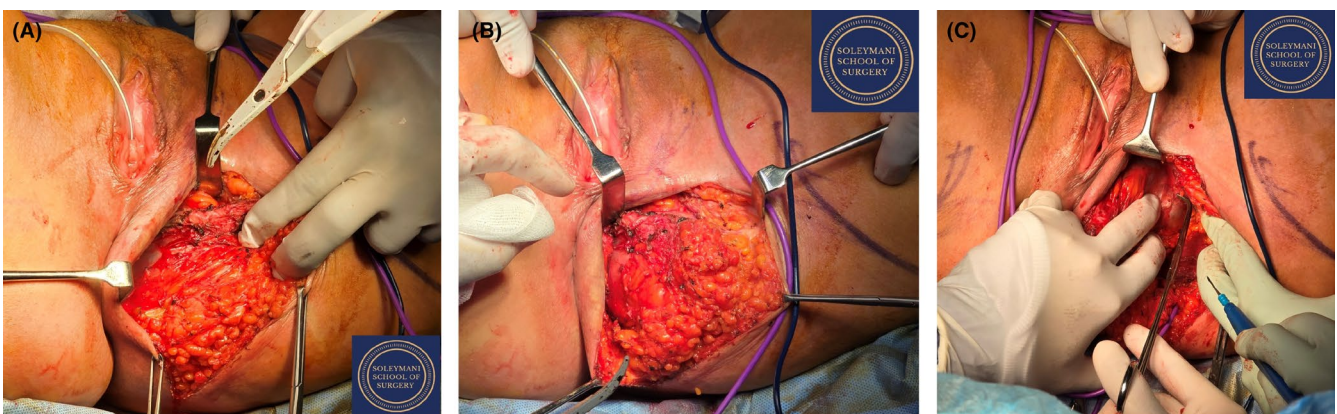


FIGURE 5 | Surgical excision of tumor and tissue margin, with careful devascularization of the tumor; (A), dissection of the overlying skin, revealing subcutaneous fat; (B), further dissection to reveal the tumor, taking care not to breach the tumor; (C), particular care was taken to preserve the pudendal nerve and to maintain good blood flow to the area.



FIGURE 6 | The mass was further excised until only the posterior wall remained to be dissected. Careful division of vessels supplying the posterior wall of the mass was carried out using LigaSure Exact.

secured in place using 2–0 Vicryl, 3–0 Monocryl, and 3–0 Vicryl interrupted sutures (Figure 14).

Two size 15 Blake drains were inserted during flap closure. This was secured using 2.0 silk suture, and topical chloramphenicol was applied.

4.10 | Step 10—Postoperative Review

The patient recovered well from the anesthetic and did well on the ward in the days following the operation. The first drain was removed 3 days postoperatively, and the second was removed 6 days postoperatively. She was on intravenous antibiotics (co-amoxiclav) for 5 days. She was discharged home 6 days postoperatively with venous thromboembolism prophylaxis (Dalteparin) and instructions to not sit or lie supine for 2 weeks. While in hospital, she only required basic analgesia (paracetamol, ibuprofen, and codeine) and was discharged on these medications to use as required.

5 | Outcome and Follow-Up

The patient was seen in clinic with the operating surgeon 4 weeks postoperatively, where she reported she was healing well with no complications. She did not report any problems with pain control following discharge. The histology report of the excised tumor reported complete removal with clear margins, with a final histological grade of 2 on the postoperative pathology report. Imaging 2 months postoperatively showed no disease recurrence.

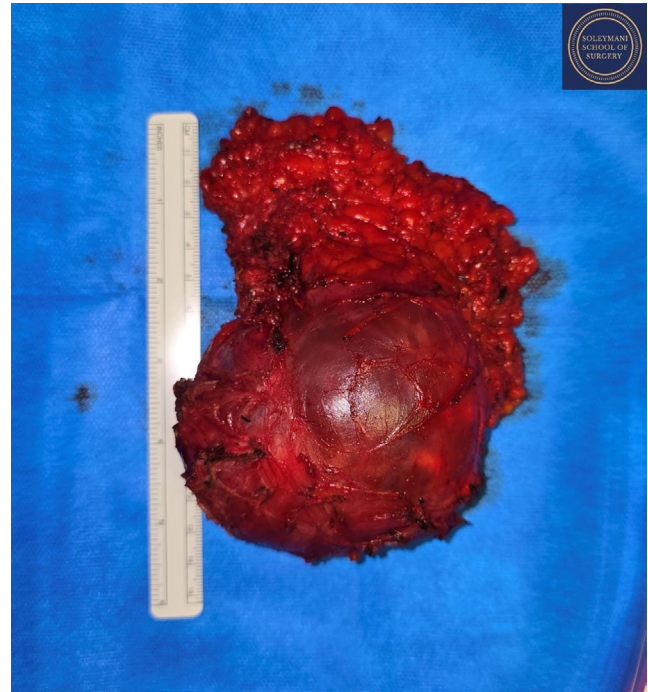


FIGURE 7 | The encapsulated tumor, measuring 8.5×7.5×6.5 cm, was removed intact and with adequate margins, pictured with a tape measure for scale.

6 | Discussion

Myxofibrosarcomas (MFS) are an uncommon pathology, accounting for approximately 5% of soft tissue sarcomas [23]. MFS usually presents as a slow-growing, painless mass. The most common locations are the extremities (77%) and the trunk (12%), followed by the head and neck region (3%) [23]. They have a median age at presentation of 64 and a slight male predominance [24]. They are a particularly rare finding when located in the IRF, first published in a case report over 20 years ago with limited follow-up or further literature since [12].

Histologically, they are characterized by the presence of myxoid and fibrous components. They were first recognized as a pathologic entity different from malignant fibrous histiocytoma by the 2002 WHO criteria [25]. There are no specific immunohistochemical markers used to diagnose MFS at present [26], though recent whole-exome sequencing reports frequent alterations in p53 signaling, and changes to cell cycle regulator genes were associated with overall poorer survival [27]. MFS lacks a pseudocapsule and typically shows an unusual infiltrative growth pattern along the fascial planes and muscle bundles. The extent of invasion is best assessed on MRI. T1- and T2-weighted MRI, with pre- and post-gadolinium imaging, should be performed to adequately assess the extent of disease [23]. Gadolinium-enhanced, fat-suppressed T1-weighted MRI may illustrate best the infiltrative growth pattern [28]. The “tail sign,” multidirectional signal spreading along the fascial plane, is a characteristic finding of MFS on MRI and can be easily overlooked [29]. This infiltrative pattern can result in anatomically deceptive margins, hence why surgical excision with a wide surgical margin is essential. The “tail sign” has been associated with a higher risk of local recurrence of disease following surgical excision by

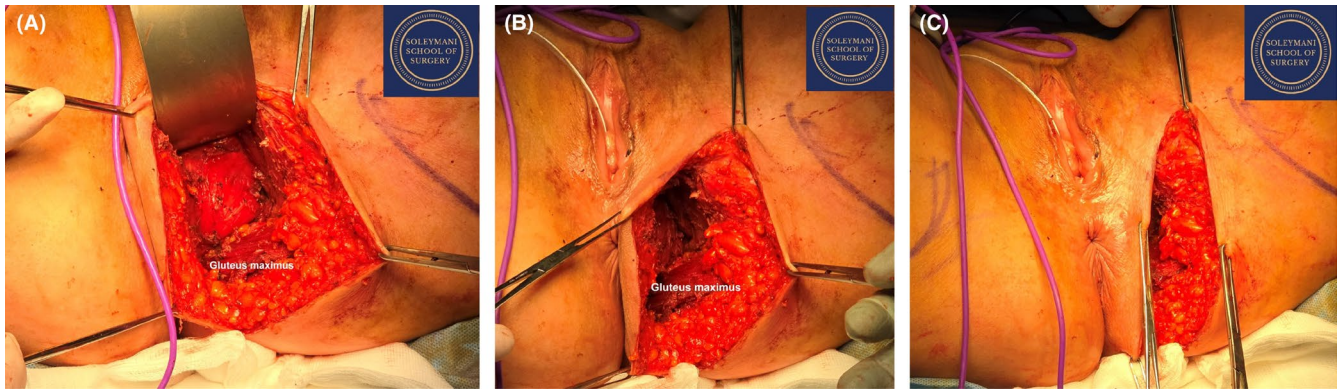


FIGURE 8 | Surgical plane following excision of tumor, with exposure of the gluteus maximus muscle; (A), the space left by removal of the tumor, pictured with retractors and the gluteus maximus labeled; (B), the space left by removal of the tumor, pictured with only the skin retracted and gluteus maximus labeled; (C), the space left by removal of the tumor with no tension on the retractors, to demonstrate the natural positioning of the remaining tissue.



FIGURE 9 | After the perforator vessels had been isolated, the flap was planned, and further incisions were made to allow for full mobilization.

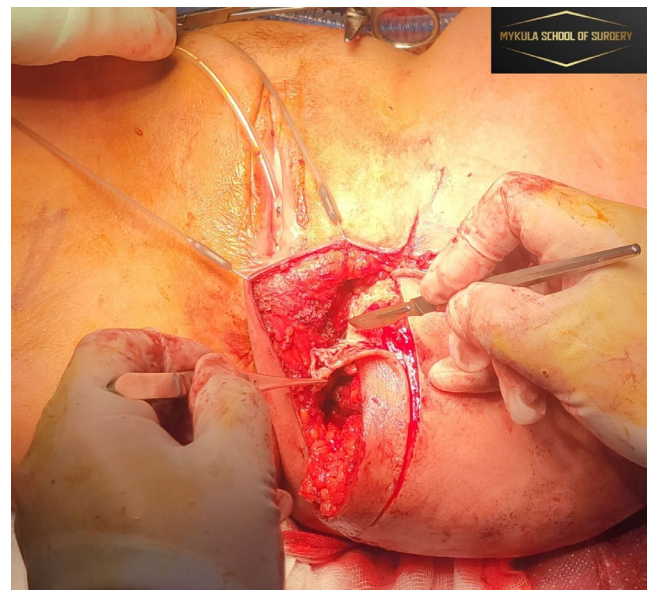


FIGURE 11 | 3–6 cm of the medial aspect of the IGAP flap was de-epithelialized anteriorly to posteriorly.

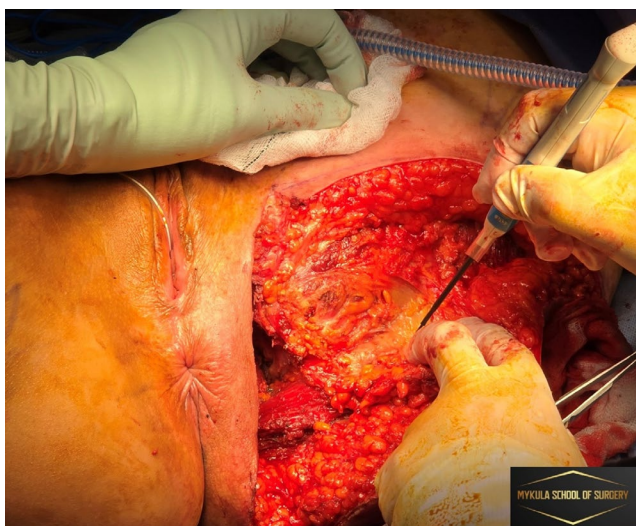


FIGURE 10 | IGAP flap divided down to the level of the gluteus maximus muscle using monopolar diathermy, to allow for mobilization.

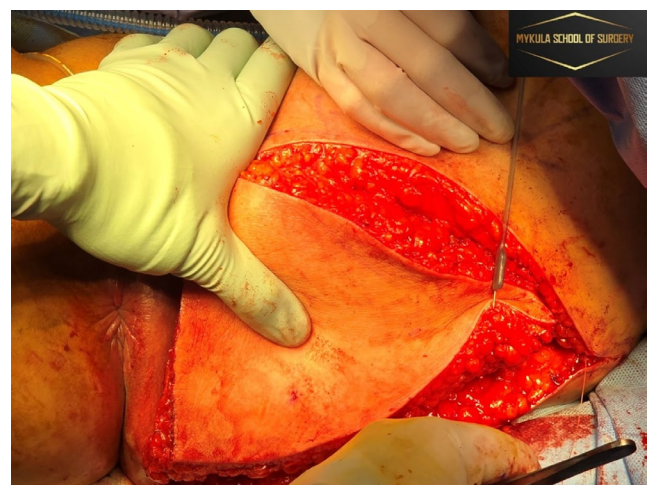


FIGURE 12 | The IGAP flap was advanced medially by 7 cm, and the de-epithelialized segment was inserted into the IRF; the lateral two skin edges were sutured.

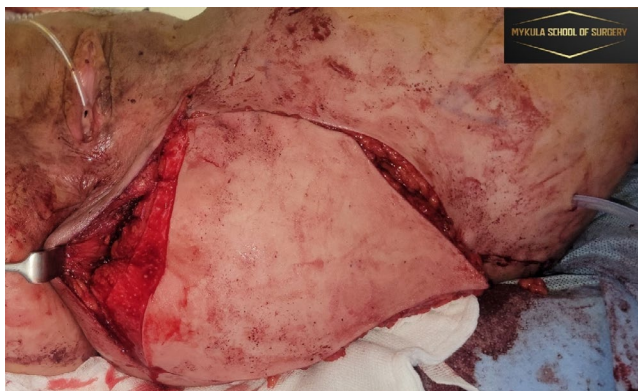


FIGURE 13 | The flap was secured using 2–0 Vicryl, 3–0 Monocryl, and 3–0 Vicryl interrupted sutures. Two size 15 Blake drains were inserted during flap closure.



FIGURE 14 | The final positioning of the flap, secured in place with interrupted sutures. Topical chloramphenicol was applied, ending the surgical procedure.

Spinnato et al. [30], who also proposed a specific MRI grading system for MFS, taking the unique radiological features into account when estimating the risk of local recurrence.

They have a low risk of distant metastases but high rates of local recurrences, up to 50%–60% [26, 31]. In up to 38% of cases, local recurrences of low-grade MFS progress to a higher histological grade, with an increase in metastatic potential [32]. Therefore, optimal surgical resection with clear margins is key. Overall survival for patients with localized MFS was reported as 61%–77% at 5 years [24, 33]. Factors associated with a poor prognosis are as follows: tumor size over 7.5cm²⁵, tumor grade [34], a high mitotic rate of more than 20 mitoses per high-powered field [31], and resection with positive/close margins [33].

Although there are no guidelines defining the optimal extent of the surgical margin in MFS, most authors suggest aiming for at least a 1–2cm margin when anatomically feasible [23]. The appropriate extent of surgical resection can be guided by the extent of soft tissue edema seen on preoperative MRI, the diameter of which is often much greater than that of the actual tumor and may delineate the infiltrative tumor edges [35].

For radical excision of IRF tumors, a perineal approach offers the most direct route for low-lying, well-circumscribed lesions, enabling sphincter preservation; however, perineal wounds carry risks of infection, delayed healing, and herniation [36]. A transgluteal approach provides a lateral corridor along the obturator internus/piriformis that avoids a contaminated perineal field and can give excellent control for lesions tracking towards the greater sciatic notch or compressing the sciatic nerve. Its drawbacks are potential injury to the sciatic and gluteal neurovascular bundles and postoperative neuropathic pain, and evidence is largely limited to case reports/series [37]. When tumors are large, malignant, or invade the levator ani-/suprlevator compartment, a combined abdomino-perineal approach—often as part of APR (abdomino-perineal resection) and ELAPE (extralevator abdominoperineal excision)—allows vascular control, wider en-bloc resection, and higher R0 rates at the cost of greater physiological insult and with risks of urinary and sexual dysfunction, as well as perineal wound complications possibly requiring flap reconstruction and carry risks of urinary/sexual dysfunction [6, 38]. The choice is therefore driven by size, cranio-caudal extent, compartmental spread, and suspected biology.

With regards to reconstructive surgery, the IGAP flap is a versatile option for perineal reconstruction, particularly following excision of IRF tumors or APR, offering reliable bulk, preservation of muscle function, and low donor-site morbidity [39, 40]. Compared with the gracilis flap, which is technically straightforward and useful for small–moderate defects but prone to atrophy and limited volume, the IGAP provides more durable tissue coverage and better contour for larger cavities [41]. The internal pudendal artery perforator (IPAP) flap offers a local fasciocutaneous alternative with minimal donor-site morbidity and shorter operative time, though its arc of rotation and bulk are limited, making it less suitable for extensive pelvic floor defects [42]. In contrast, the vertical rectus abdominis myocutaneous (VRAM) flap provides robust, well-vascularized tissue ideal for irradiated or extensive defects, but at the expense of significant donor-site morbidity, abdominal wall weakness, and potential hernia formation [42, 43]. Thus, IGAP is often favored when durable volume and reliable vascularity are required without compromising muscle, while gracilis and IPAP serve smaller or superficial reconstructions, and VRAM is reserved for the most extensive or previously irradiated perineal wounds.

While there is no high-quality evidence supporting the use of radiotherapy for MFS specifically, radiation is considered an effective adjuvant treatment, based on randomized controlled trials looking at soft tissue sarcomas [44, 45]. Chemotherapy is not routinely administered in the treatment of MFS, as there are no studies suggesting benefit [23, 24]. Immunotherapy options may be explored in the future, due to the presence of a “T-cell inflamed” tumor microenvironment [46].

7 | Conclusion

Surgical resection with a clear margin is the cornerstone of treatment for MFS. We present the exceptionally rare case of a myxofibrosarcoma occurring in the IRF. This patient underwent multidisciplinary surgery by the Gynecology Oncology, Orthopedics, and Plastics teams, in which complete

resection with clear margins was achieved, with no complications or loss of function. We describe a 10-step approach for the perineal approach of the IRF tumor, with IGAP V-Y flap reconstruction.

Author Contributions

Amy Whitmore: writing – original draft. **Sabina Ioana Nistor:** writing – original draft, writing – review and editing. **Muhammad Ather Siddiqi:** writing – review and editing. **Roman Mykula:** writing – review and editing. **Hooman Soleymani Majd:** writing – original draft, review, editing and supervision.

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Ethics Statement

No conflict of interest identified. The patient gave written consent for this case report to be published. No permission was required to reproduce any sources.

Data Availability Statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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