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Ischiorectal Fossa Mass: Angioleiomyoma Workup and Treatment

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Background	Angioleiomyoma, a benign pericytic neoplasm derived from vascular smooth muscle, typically presents as a subcutaneous nodule in the lower extremities during the fourth to sixth decades of life. Its occurrence in the ischiorectal fossa is rare, leading to a paucity of data in colorectal surgical literature and presenting diagnostic challenges due to nonspecific features.
Summary	We report the case of a 58-year-old male who presented with a large, symptomatic mass in the right perianal soft tissues causing discomfort upon sitting. Cross-sectional imaging (CT and MRI) revealed a well-defined, lobulated mass within the ischiorectal fossa appearing to originate from the pelvic floor musculature. Preoperative percutaneous biopsy confirmed the diagnosis of angioleiomyoma, and endoscopy excluded intraluminal pathology. The patient underwent successful trans-perineal surgical excision of the mass, which necessitated resection of a small portion of the adjacent external anal sphincter fibers. Final histopathology confirmed angioleiomyoma.
Conclusion	Angioleiomyomas in this unusual location can be effectively managed with complete surgical excision, leading to symptom resolution. However, this case also underscores the critical importance of a comprehensive preoperative evaluation, including imaging and tissue biopsy, to accurately differentiate these benign lesions from malignant tumors of the perirectal region, thereby guiding appropriate surgical strategy and management.
Key Words	angioleiomyoma; ischiorectal fossa; perirectal mass; surgical excision

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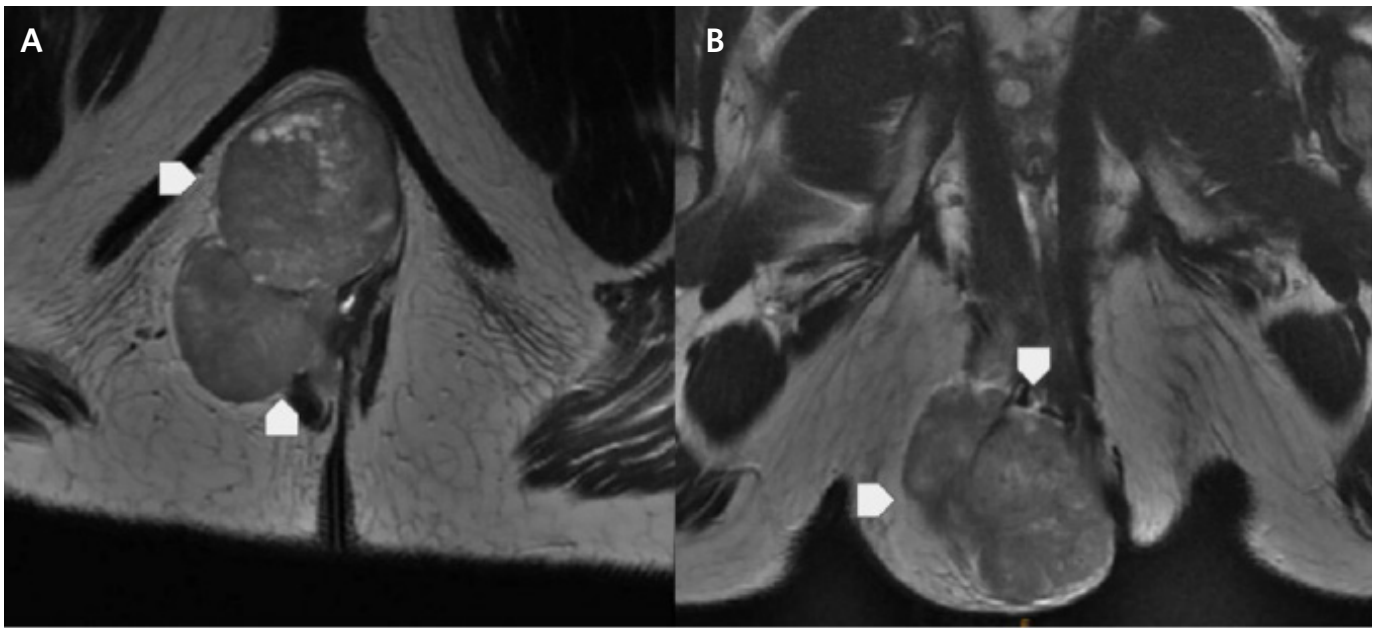
Case Description

Angioleiomyomas are benign pericytic neoplasms originating from the tunica media smooth muscle layer of blood vessels, typically small veins or venules.[1] Grossly, they commonly manifest as solitary, well-circumscribed, firm, slow-growing, subcutaneous nodules. While these lesions can occur at any age, they most frequently present during the fourth to sixth decades of life, with a marked predilection for the lower extremities (50-70% of cases).[2,3] The most characteristic symptom, when present, is localized pain without distal neurovascular involvement, though many angioleiomyomas remain asymptomatic.[4] Owing to their rarity and nonspecific clinical features, the diagnosis typically relies on a combination of clinical suspicion and definitive histopathological evaluation following biopsy or excision. Given the paucity of data in the existing literature, particularly regarding unusual locations, we present a rare case of angioleiomyoma arising within the ischiorectal fossa.

A 58-year-old male with a past medical history of hypertension, hyperlipidemia, and hypothyroidism presented for evaluation of progressively enlarging mass in the right inner buttock over the previous three years. His primary symptom was discomfort pronounced when sitting. A screening colonoscopy performed eight years prior was unremarkable, and hisfamily and surgical histories were noncontributory. Physical examination revealed a fist-sized, firm, mobile, and nontender mass located within the soft tissues lateral to the anal sphincter complex and medial to the right ischial tuberosity, seemingly contained within the ischiorectal fossa. The overlying skin and subcutaneous tissues appeared normal. Anal sphincter tone was intact, and a digital rectal exam revealed no intraluminal mass.

Initial workup, initiated by the patient's primary care physician four months prior to presentation, included a CT scan of the abdomen and pelvis. This study identified a 7.5 × 4.4 × 9.8 cm lobulated soft tissue mass exhibiting heterogeneous density within the right ischiorectal fossa. Subsequent MRI of the pelvis provided further characterization, demonstrating a multilobulated, smoothly marginated, enhancing mass arising appearing to arise from the undersurface of the pelvic floor and extending inferiorly through the right ischiorectal fossa (Figure 1). There was no obvious invasion into the adjacent anorectal structures or involvement of pelvic osseous structures, and no significant lymphadenopathy was noted. Anoscopy and colonoscopy were performed, confirming normal anorectal mucosa throughout without evidence of intraluminal lesions.

Figure 1. MRI of Ischiorectal Fossa Angioleiomyoma. Published with Permission



T2-weighted MRI of the pelvis. ***(A)*** *Axial and* ***(B)*** *Coronal views demonstrating a large, well-circumscribed, encapsulated mass within the right ischiorectal fossa. The mass exerts significant mass effect, displacing the anorectal structures medially, but shows no definitive evidence of local invasion into adjacent muscle or bone.*

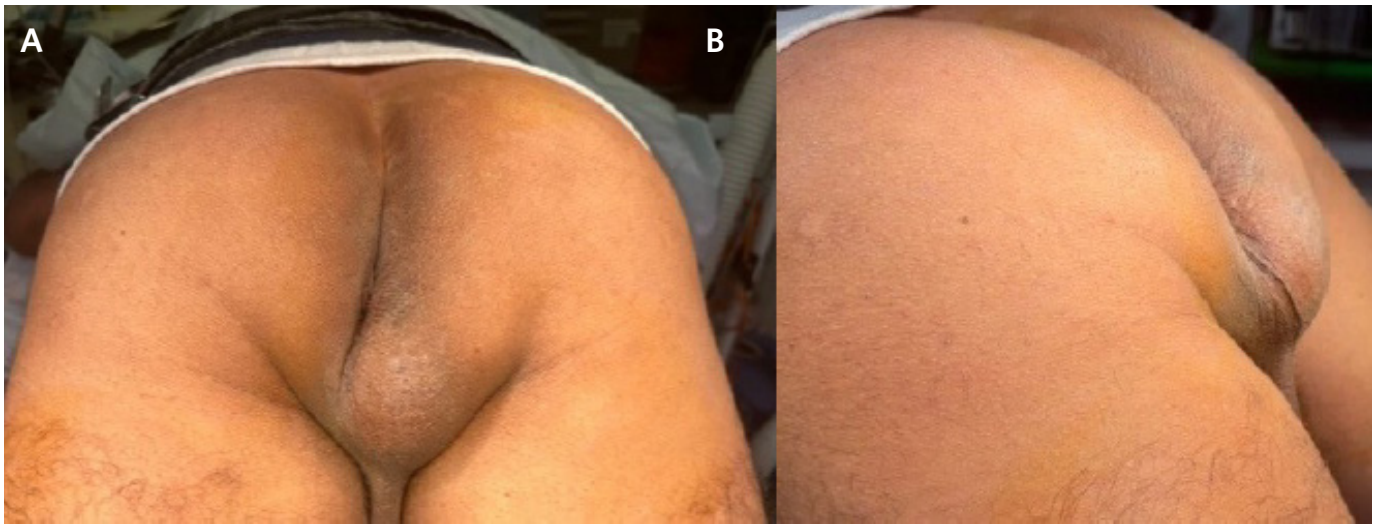
To rule out malignancy prior to surgical planning, a percutaneous core needle biopsy was performed. Histopathological examination revealed spindle cells positive for smooth muscle actin (SMA), exhibiting features consistent with smooth muscle differentiation, accompanied by a prominent vascular component. This morphology confirmed the diagnosis of angioleiomyoma. Given the benign nature of the lesion, trans-perineal excision was planned.

The patient underwent a preoperative cathartic and antibiotic bowel preparation as a precaution against inadvertent rectal entry during the operation. Following induction of general anesthesia and administration of pre-incisional antibiotics, he was positioned prone (Figure 2). A perianal incision provided access to the ischiorectal fossa, revealing a well-encapsulated, well-circumscribed, multilobulated mobile mass consistent with imaging findings (Figure 3). The mass possessed a thick-appearing capsule and was situated primarily within the ischiorectal fat pad. Dissection proceeded readily, identifying the superior-medial aspect of the mass originating from the undersurface of the levator ani and external anal sphincter (EAS) complex. A small portion of the tumor was noted to extend through a defect within the EAS, abutting the internal anal sphincter (IAS). Careful dissection preserved the IAS and the overlying intact rectal mucosa, confirmed visually with intraoperative anoscopy. A small cuff of EAS fibers directly involved by the tumor extension was necessarily excised en bloc with the specimen (Figure 3). Following complete removal of the mass, the defect in the EAS was repaired, and the ischiorectal space and skin were closed in layers. Final anoscopy and proctoscopy confirmed anorectal mucosal integrity without evidence of bleeding.

Surgical pathology confirmed the diagnosis of angioleiomyoma. Examination revealed a proliferation of spindle cells demonstrating smooth muscle features and prominent vascularity, without significant cellular atypia or abnormal mitotic activity. Immunohistochemical staining showed positivity for SMA and negativity for S100, CD34, and pan-keratin (AE1/AE3).

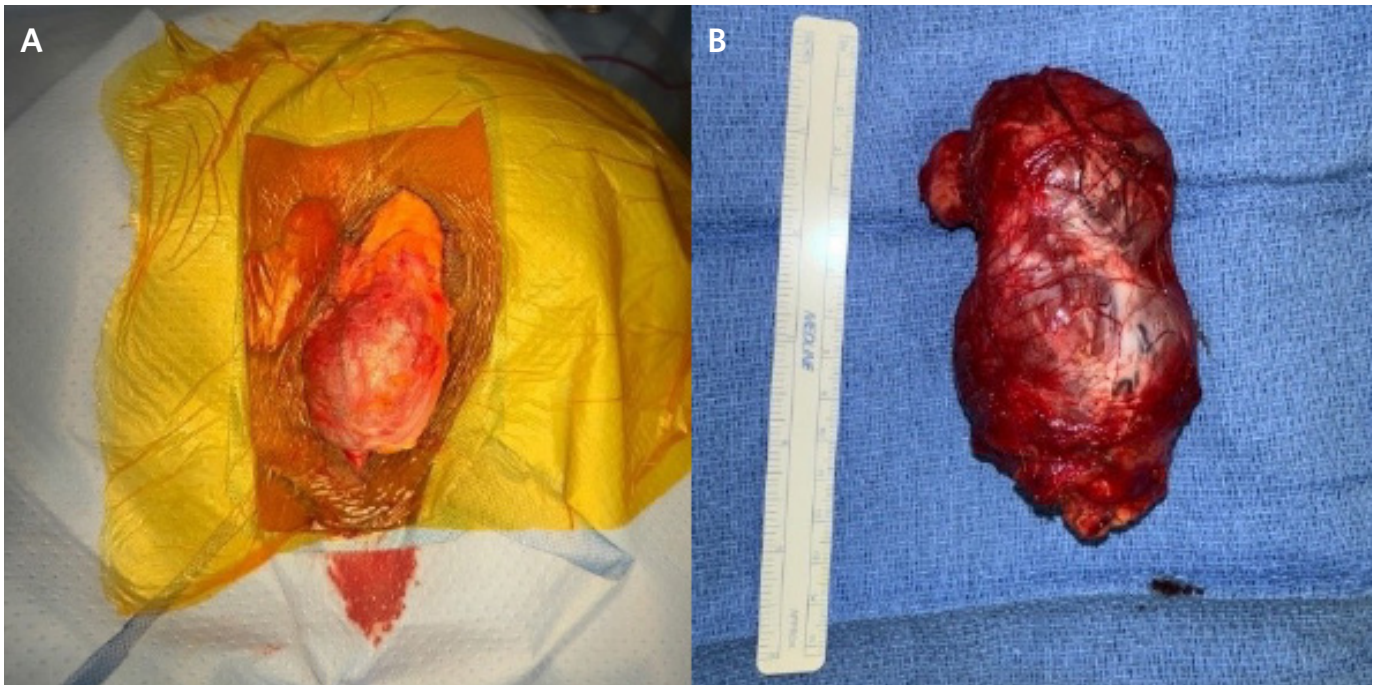
The patient's postoperative course was notable only for transient urinary retention, which resolved with temporary Foley catheterization and tamsulosin. He was discharged on the first postoperative day, with the Foley catheter subsequently removed in the outpatient setting. At his one-month follow-up visit, the patient reported complete resolution of his presenting symptom (discomfort with sitting) and described normal bowel habits and sphincter function, with no fecal incontinence.

Figure 2. Preoperative Clinical Presentation. Published with Permission



Clinical photographs of the perianal region prior to surgery. ***(A)*** *Posterior view illustrating the location of the mass in the right ischiorectal fossa (anterolateral to the anal verge, medial to the ischial tuberosity). Note the subtle overlying skin changes attributed to chronic friction.* ***(B)*** *Lateral view demonstrating the prominent bulge created by the underlying mass, causing deviation of the median raphe to the left.*

Figure 3. Intraoperative Findings and Gross Specimen. Published with Permission



Intraoperative photographs from the trans-perineal excision. ***(A)*** *Initial dissection reveals the encapsulated, well-circumscribed, multilobulated soft tissue mass situated within the ischiorectal fat pad.* ***(B)*** *The completely excised surgical specimen, highlighting its distinct lobulations and firm, fibrous/muscular appearance with noticeable surface vascularity.*

Discussion

This case highlights the diagnostic evaluation and successful surgical management of an uncommon tumor, an angioleiomyoma, presenting in the rare location of the ischiorectal fossa. Masses arising in this anatomical region necessitate a thorough and systematic workup. Key components of this evaluation include precise anatomical delineation and assessment of malignant potential. Imaging modalities such as CT and particularly MRI are crucial for defining the extent of the mass and its relationship to critical adjacent structures, including the pelvic floor musculature, anorectal sphincter complex, and pelvic organs. Functional studies like defecography may occasionally provide additional information. Determining whether the lesion is benign or malignant is paramount and typically requires tissue sampling, either via image-guided biopsy or endoscopic biopsy if the lesion involves the colorectal lumen. This distinction fundamentally dictates the therapeutic strategy: benign lesions, such as the angioleiomyoma in this report, are often amenable to complete local excision, whereas malignant tumors typically warrant more aggressive multimodal therapy, potentially including radical surgery (e.g., abdominoperineal resection) and adjuvant treatments.

Angioleiomyomas belong to the category of benign pericytic (perivascular) neoplasms, believed to originate from the smooth muscle cells normally surrounding blood vessels. Histologically, they are characterized by well-differentiated spindle cells with smooth muscle features intermingled with prominent, often thick-walled, vascular channels. Immunohistochemistry typically reveals strong positivity for SMA.[7] While frequently asymptomatic and discovered incidentally, angioleiomyomas can cause symptoms related to their size and location, as exemplified by the positional discomfort experienced by our patient due to mass effect within the confined ischiorectal space.

The differential diagnosis for a soft tissue mass in the ischiorectal fossa is broad. Within the spectrum of pericytic and spindle cell tumors, considerations include glomus tumors, schwannomas (also classified as neurogenic tumors), and malignant entities like synovial sarcoma; specific immunohistochemical panels and radiologic characteristics aid in distinguishing these.[7] More broadly, the differential for presacral and perirectal masses encompasses primary malignancies (adenocarcinoma, squamous cell carcinoma, lymphoma, gastrointestinal stromal tumor [GIST], leiomyosarcoma, neuroendocrine tumor, melano-

ma), developmental lesions (e.g., tailgut cysts), other neurogenic tumors, and primary osseous tumors arising from the sacrococcygeal spine.[8]

Given the patient's history of hepatic hemangiomas and skin tags, neurocutaneous lesions, such as tuberous sclerosis (TSC), were considered. However, TSC was deemed less likely due to the absence of pathognomonic features such as characteristic cutaneous lesions (hypopigmented macules, facial angiofibromas, periungual fibromas) and the presence of an ischiorectal angioleiomyoma rather than the classic renal angiomyolipoma associated with TSC. The patient's age and normal renal function further lowered the suspicion for TSC. Ultimately, the percutaneous biopsy proved indispensable in this case, confirming the diagnosis of angioleiomyoma and reliably excluding the numerous malignant possibilities within the differential diagnosis.

Summary

This report details the successful management of an angioleiomyoma, a rare benign pathological entity, arising in the uncommon location of the ischiorectal fossa. To our knowledge, few such cases have been documented. Our experience underscores that once malignancy is excluded through comprehensive diagnostic evaluation, including high-resolution imaging like MRI and definitive histological confirmation via biopsy, these lesions can be effectively treated with complete surgical excision, leading to favorable outcomes and resolution of local compressive symptoms.

Lessons Learned

Crucially, the clinical presentation of an ischiorectal fossa mass can mimic a wide spectrum of both benign and malignant pathologies originating in the perirectal and presacral regions. Therefore, a prudent and thorough workup is imperative to establish an accurate diagnosis before proceeding with surgical intervention, primarily focused on excluding malignancy. Surgical excision should be considered for symptomatic lesions.

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