Fistula-in-ano is a common perianal condition that is usually attributed to cryptoglandular disease. In contrast, presacral cysts are a rare finding, reported to occur in 1 of 40,000 general hospital admissions. Presacral cysts may manifest as pelvic pain, pelvic organ dysfunction, or neurological dysfunction. Perianal suppuration can occur, but this is uncommon. We report a case of an ectopic focus of prostatic tissue presenting as a fistulizing presacral cyst. To our knowledge, this is the first reported case of ectopic prostatic tissue presenting as a fistula-in-ano.

**Case report**

A 40-year-old man presented to the emergency department because of a recurrent right perianal abscess. He reported having several such abscesses treated by incision and drainage at outside facilities over the preceding years. He also had a recurrent pilonidal cyst, which required multiple recent surgeries. He had no other significant medical or surgical history and was otherwise healthy.

Physical examination revealed multiple scars over the inferior sacrum and coccyx, consistent with pilonidal surgeries, and well-healed scars on the right lateral perianal skin, consistent with multiple drainage procedures for recurrent perianal abscesses. On digital rectal examination, there was exquisite tenderness of the right lower rectal vault with extrinsic compression at the right posterolateral quadrant. There was no drainage, fluctuance, erythema, or swelling of the perianal skin, and the rest of the examination was unremarkable.

Because the patient’s pain appeared to be out of proportion to the physical findings, a computed tomography (CT) scan was performed. This revealed a 3.6 x 5.4-cm presacral fluid collection tracking to the right perianal space (Figure 1). He was taken to the operating room for examination under anesthesia, and the right perianal abscess was drained through the existing scar. His immediate postopera-
tive course was uneventful, and his symptoms completely resolved by his first follow-up examination.

Several months later, the patient had multiple recurrences, which prompted numerous radiological studies and operative interventions. Fistulograms through a single external opening revealed a presacral cavity with no internal opening. The cavity measured 1 to 2 cm in the anteroposterior axis. Additional focal cavitary areas were connected to the main cavity by 3- to 5-mm sinus tracts (Figure 2). Many other plain radiographic studies, including an intravenous pyelogram, were performed in response to symptoms reported during emergency department visits; all studies were normal.

Because of his recurrent symptoms and despite normal imaging studies, the patient was again examined under anesthesia. A lacrimal probe examination and injection of methylene blue confirmed that the presacral cavity had no internal opening. Curettage of the fistula tract to the deep postanal space and instillation of fibrin glue were performed. A pathology examination of tissue recovered from the tract revealed only chronic inflammation consistent with a fistula and no cancerous or ectopic cells. The patient’s symptoms were dramatically relieved, and a CT scan revealed resolution of acute inflammation.

The patient was subsequently lost to follow-up until his symptoms recurred approximately 1.5 years later. A repeat CT scan revealed recurrence, and the patient had increased pain and drainage from the presacral cavity. He was taken to the operating room, where an injection of

![Figure 1](image1.png)  
**Figure 1**—CT scans showing the presacral cyst (A) and its fistula tract (B).

![Figure 2](image2.png)  
**Figure 2**—Fistulogram showing anteroposterior (A) and lateral (B) projections.
methylene blue revealed a posterior internal opening 1 cm proximal to the dentate line. Probing found a transsphincteric complex fistula leading to the presacral space. The abscess was incised and drained, and the internal opening was closed. Instillation of fibrin glue and placement of a Malecot® drain into the right-sided abscess were undertaken. The patient healed well after the drain was removed. He was followed up a couple of times and showed significant improvement of his symptoms and complete resolution of the drainage. He was ultimately discharged from surgical follow-up.

Approximately 1 year later, the patient underwent multiple studies to evaluate new symptoms, consisting chiefly of a change in how he characterized his pain. He described it as constant but not as severe as it was previously. These studies showed near resolution of the posterior cavity with improvement in the inflammatory process; however, a sinus tract into the right perianal space and a right perirectal mass-like lesion with compression of the rectum were partially visualized.

Due to the patient’s multiple recurrences and history of pilonidal disease, magnetic resonance imaging (MRI) was performed, which showed a 3.4 x 4.6-cm cystic mass leading to the ischiorectal space (Figure 3). The mass extended anteriorly but did not involve the prostate or seminal vesicles. Upper and lower endoscopies to evaluate other gastrointestinal symptoms were noncontributory at the time. Since both of these studies were negative for ulcer disease or inflammatory bowel disease, a diagnosis of presacral cyst was made.

The patient was brought to the operating room, where coccygectomy and dissection of the levators were performed via a posterior midline incision. This revealed a cystic structure approximately 1 cm inferior to the level of the coccyx and extending 2 cm superior to the coccyx. It was not attached to the posterior anal canal or rectum and was resected in its entirety. Gross inspection showed the cyst was smooth-walled with thick, tan, opaque fluid; two smaller associated cysts were found. There was a tract leading to the right ischiorectal abscess cavity, consistent with the radiologic findings. No tracts were found extending anteriorly to the prostate, and the anterior planes showed no evidence of acute or chronic inflammation. Histological findings revealed prostatic tissue in the cyst via hematoxylin and eosin staining; this was confirmed by special stains, including prostatic-speciﬁc antigen (PSA) and prostatic acid phosphatase (PAP; Figure 4).

Discussion
Although there are other reports of ectopic prostatic tissue, most describe patients who were asymptomatic and

Figure 3—T1-weighted (A) and T2-weighted (B) MRIs showing the cyst.
whose tissue was found on pathological examination of the surgical specimens. A few reports involve distant organs, such as the spleen or lungs, but the most common area of involvement is the posterior urethral region. When the condition is symptomatic, it almost always presents as gross, painless hematuria.\textsuperscript{2-4} We found four cases describing ectopic prostatic tissue involving the anorectum; two were symptomatic like our patient.\textsuperscript{8-12} In 2001, Fulton and colleagues reported the case of a 78-year-old man presenting with a 2.5-cm retrorectal mass that caused obstructive bowel symptoms.\textsuperscript{10} Like our patient, he underwent MRI, which showed the nodule. It was excised and found to have prostatic tissue with surrounding stromal tissue. Overall, this case was very similar to ours and included multiple cysts. Histological and immunohistochemical analysis showed the nodule was strongly positive for PSA. Immunoperoxidase staining confirmed prostatic differentiation. The unusual location was presumed to be related to the normal embryologic origins of the rectum, bladder, and prostate; these structures are contiguous during early gestation.

In 1913, Lowsley described embryogenesis of the prostate. The rectum and bladder are derivatives of the endodermal cloaca. At 5 weeks gestation, the two organs are divided by a caudal mesodermal extension. At around week 12, there is initial development of what is to become the prostate. The anterior organ further develops by extending tubular tissue that encircles the base of the bladder and urethra to form the prostate. More glands develop at 18 weeks, forming the urethral glands of Littre, the subcervical glands of Albarran, and the subtrigonal glands.\textsuperscript{13}

Fulton and colleagues postulate that prostatic tissue can follow the posterior division and rest in the posterior compartment, forming the ectopic organ; thus, the authors feel the term “ectopic prostate” more accurately describes the lesion based on its embryologic origins.\textsuperscript{10} This theory most likely also represents the process in our patient.

Tekin and colleagues reported the case of a 59-year-old man who had a bleeding, 2.5-cm sessile anal polyp, which was excised and confirmed to contain ectopic

![Figure 4](image-url)
prostatic tissue on histological and immunohistochemical studies.\(^9\) Smooth muscle and glands were present, consistent with benign nodular hyperplasia. The authors discussed the alternative theory of tissue seeding from previous surgery or biopsy. This is unlikely to have been the case with our patient, since he reported no manipulation in the region of the prostate and had no intraoperative or radiologic findings that would support this theory.

Morgan reported the case of an elderly man with paraplegia who was found to have prostatic tissue at the anus upon debridement of a decubitus ulcer.\(^11\) The patient had no history of urological surgery or other possible events to account for the tissue’s presence. The pathology techniques that identified the prostatic tissue in this man’s case were similar to those conducted by our pathologists.

Gledhill reported the case of a patient who underwent a proctocolectomy for ulcerative colitis.\(^12\) At operation, an asymptomatic presacral mass was found in the perirectal fat. Histologic examination revealed prostatic tissue. Gledhill’s case supports the theory of tissue organization failure, which proposes that there is a lack of differentiation of tissues in the pelvic region into their typical type. This differs from mature tissues migrating into abnormal areas.

In 2005, a report was published summarizing the literature to date regarding ectopic prostatic tissue associated with the bladder.\(^14\) Only 32 cases are identified in the English-language literature, and patients reportedly presented with gross or microscopic hematuria. This paper noted that ectopic tissue is most frequently found in the male urethra and infrequently found in the bladder neck, trigone, and interureteral ridges.

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**Ectopic prostatic tissue in the presacral space can masquerade as other common diseases of the anus and rectum.**

Based on the literature, the mechanism of ectopic tissue has three possible explanations. First, ectopic tissue may represent persistent tissues that typically regress during embryological development. Second, ectopic tissues may represent metaplastic changes of transitional epithelium due to inflammation or other etiologies; these may be more applicable to patients with urological manifestations. Third, ectopic tissue may arise from abnormal migration of undifferentiated cells during embryogenesis with hormonal stimulation as an adult. This last theory would be the most likely explanation for our patient, because he had tissue clearly distant from the expected sites of ectopia. Other examples supporting this third theory include prostatic tissue found in the spleen, pericolic fat, uterus, cervix, and anus.

Ectopic prostate is clinically important because it can cause several different symptoms. If found in the lower genitourinary system, it typically causes hematuria.\(^7\) When it occurs within the anorectum, it can cause obstructive symptoms, bleeding, or infection.\(^9,10\) Although no evidence of adenocarcinoma was found in
our patient, there is one report of adenocarcinoma arising from ectopic prostatic tissue. As previous authors concluded in other cases, we think that our patient's prostatic tissue was present in the anus or rectum due to the common embryologic origin of the lower urinary tract, anus, and rectum. This theory is supported by the intraoperative and radiologic findings of a distinct, separate lesion.

Conclusion

Ectopic prostatic tissue in the presacral space is extremely rare and can masquerade as other common diseases of the anus and rectum. It is difficult to assess the symptomology and likelihood of malignant changes because of the rarity of this condition. MRI is likely the best study to identify these lesions. Traditional radiology studies can be useful in planning the surgical approach but are not useful in predicting the presence of prostatic tissue. Special immunohistochemical stains must be used if there is suspicion of aberrant tissue. Complete excision of the tissue may prevent recurrence.

References