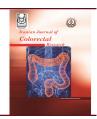
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Perineal Ectopic Prostate Tissue: A Rare Case and Its Clinical Implications

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Abstract

Ectopic prostate tissue is a rare condition characterized by the presence of prostate glands outside their typical anatomical location, rather than the entire organ being displaced. While it is most commonly observed in the lower male genitourinary system, it has also been reported in various other sites, including the pericolic fat, the female genitourinary system, and even the spleen. The most common location is the urinary bladder trigone, though it can also appear in the periurethral area or at the bladder neck. Here, we present a case involving perineal ectopic prostate tissue that manifested as a significant mass. A 52-year-old man visited a colorectal clinic due to a bulging, mass-like lesion in the perineal region. Perineal soft tissue sonography identified a solid-cystic mass, while pelvic magnetic resonance imaging (MRI) revealed a large solid-cystic mass located posterior to the sacrum, beneath the skin. Physical examination demonstrated a palpable, large perineal mass extending from the inferior border of the scrotum to the anterolateral aspect of the anus. Laboratory data were within normal limits. Both tru-cut biopsy and fine-needle aspiration (FNA) indicated a bloody, low-cellularity specimen with no malignant cells. Subsequently, the entire mass was excised via a transperineal approach. Pathological and immunohistochemical studies confirmed the presence of ectopic prostate tissue without any signs of malignancy. Consequently, ectopic prostate tissue should be considered in the differential diagnosis of any perineal mass, and surgical excision is recommended due to the potential risk of malignancy.

Keywords: Prostatic Neoplasm, Perineum, Tissue, Ectopic, Urogenital System

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Introduction

Ectopic prostate tissue, located distal to the apex of the prostate, is a rare condition that has been reported in various intra- and extragenitourinary sites (1). Prostatic ectopia does not refer to the misplacement of the entire organ but rather to a

scattered group of prostate glands (2). This condition is typically found in the lower male genitourinary system; however, it can also appear in the pericolic fat, female genitourinary system, anal canal, retroperitoneum, and spleen (3). Additionally, there have been reports of rare cases occurring in the epididymis, penis, and even the seminal vesicle (4).

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The most common location is the trigone of the urinary bladder, with rare occurrences in the periurethral area or bladder neck (5, 6). Ectopic prostate tissue has typically presented as a single lesion in previous studies; however, one case report revealed multiple lesions in the pelvis (7). The suspected etiologies of prostatic ectopia include the persistence of prostatic tissue during embryogenesis, metaplasia of the urinary epithelium due to chronic inflammation, and the migration of prostatic stem cells (8-10). Among various imaging modalities, magnetic resonance imaging (MRI) is highly sensitive for detecting ectopic prostate tissue. This condition can result in elevated levels of prostate-specific antigen (PSA) even after a radical prostatectomy. Ectopic prostate tissue should be considered in the differential diagnosis of any palpable perineal mass detected during a digital rectal examination. We report a case of perineal ectopic prostate tissue presenting as a large mass.

Case Presentation

A 52-year-old man presented to the colorectal surgery department at Motahari Clinic, Shiraz University of Medical Sciences, Shiraz, Iran, in January 2024, with a gradually enlarging perineal mass that had been growing over the past year,

with a rapid increase in size over the last month. A perineal soft tissue sonography performed at another center suggested a differential diagnosis of sarcoma versus squamous cell carcinoma (SCC), presenting as a solid-cystic mass. The patient's medical history was unremarkable, with no current medication use, except for episodes of constipation and urinary retention several years ago. Physical examination revealed a large, firm, and palpable mass in the perineum, extending from the inferior border of the scrotum to the anterolateral aspect of the anus. The patient did not report any pain or urinary issues. Laboratory studies were within normal limits.

A Tru-cut biopsy performed in the office yielded a bloody, low-cellularity sample, which was sent for cytological analysis. No malignant cells were detected. A fine-needle aspiration (FNA) guided by sonography yielded the same result. A pelvic MRI, conducted with and without contrast, revealed a 13×12×6 cm solid-cystic mass in the perineal area, located posterior to the sacral sac and just beneath the skin. The mass contained cystic components and exhibited heterogeneous enhancement in the solid parts, with some areas of signal void, highly suggestive of an infiltrative mass that requires correlation with biopsy results. The differential diagnosis included sarcoma versus SCC (Figure 1).

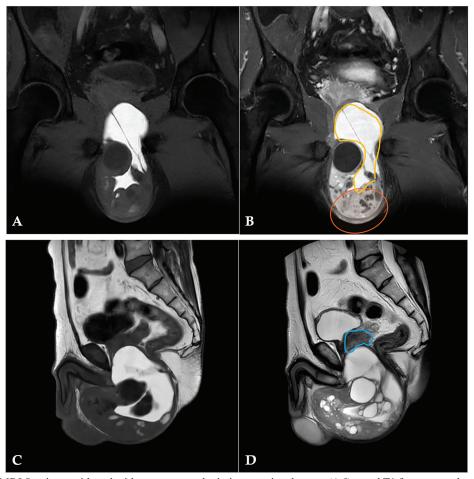


Figure 1: Pelvic MRI Sections, with and without contrast, depicting a perineal mass: A) Coronal T1 fat-saturated pre-contrast image; B) Coronal T1 fat-saturated post-contrast enhanced image; C) Sagittal T2-weighted image; and D) Sagittal T1-weighted image of the pelvic cavity. The red outline indicates the solid enhancing component, the yellow outline highlights the blood-filled cystic component, and the blue outline denotes the prostate.

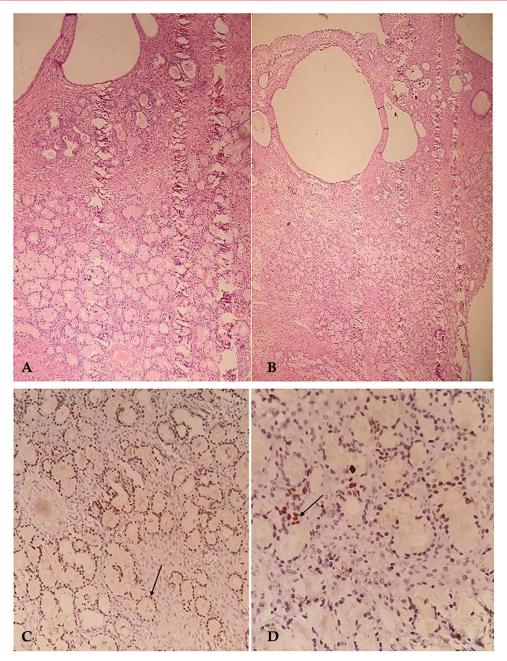


Figure 2: Histologic sections from a perineal lesion: A) Hematoxylin and eosin (H&E) stain at 100× magnification reveals mucin-producing acini and dilated ducts with calcification, resembling normal prostate tissue; B) 400× magnification of the same area; C) Immunohistochemistry (IHC) stain for NKX3, which is positive in the epithelial lining, confirming prostatic origin (arrow); and D) IHC stain for P63, which is positive in the basal cells of this benign ectopic prostatic tissue (arrow).

The entire mass was resected using a transperineal approach. The size of the mass was approximately consistent with the findings reported in the MRI. Gross examination revealed that the mass was firm, elliptical, and well-circumscribed. Pathological and immunohistochemical evaluations confirmed the presence of ectopic prostatic tissue, with no evidence of malignancy (Figure 2). The postoperative course was uneventful, and the patient was subsequently discharged.

The study protocol was approved by the Ethics Committee of Shiraz University of Medical Sciences (code: IR.SUMS.REC.1403.415). Written informed consent was obtained from the patient who agreed to participate in this study.

Discussion

Ectopic prostatic tissue can be found throughout the body, most commonly in the lower genitourinary tract of males, including the urethra, seminal vesicles, epididymis, testis, and urinary bladder (11, 12). The presence of ectopic prostate tissue in various locations beyond the male genitourinary system presents a unique diagnostic and clinical challenge. It has also been encountered in the uterus, cervix, and vagina (13). Additionally, involvement of the retrovesical space, spleen, rectum, anal canal, or pericolic fat has been described (14). While isolated ectopic prostate tissue has been reported in the bladder trigone, pericolic fat, epididymis,

and even the female reproductive system, the perineal manifestation of ectopic prostate tissue, as observed in this case, represents an exceptionally rare occurrence.

Patients typically present with lower urinary tract symptoms, such as hematuria, or obstructive symptoms, including frequency and retention (15). Other symptoms may vary depending on the location of the involvement. Unlike previously described cases, where ectopic prostate tissue was largely incidental or associated with localized urinary symptoms, this patient exhibited a progressively enlarging mass over the course of a year, raising substantial concerns regarding malignancy. The differentiation between benign ectopic prostate tissue and more aggressive pathologies, such as sarcoma or SCC presented a significant challenge in this case, further underscoring its uniqueness.

The exact etiology of ectopic prostatic tissue remains unclear. While a vestigial remnant from embryogenesis is considered a possibility for the prostatic urethra and trigone, metaplasia resulting from chronic inflammation, aberrant embryogenesis due to the migration or misplacement of the prostate gland, and the seeding of viable tissue to the rectum during surgery or biopsy are also mentioned as potential etiologies for other sites (7, 14, 15).

A major distinguishing factor in this case was the confirmation of the origin of the prostate tissue through immunohistochemical staining, specifically the positivity of NKX3 in epithelial cells and P63 in basal cells. These findings were critical, as ectopic prostate tissue histologically resembles normal prostate glands, making its identification outside the expected anatomic location particularly challenging. This case highlights the necessity of immunohistochemical markers in differentiating ectopic prostate tissue from other benign or malignant masses, thereby ensuring accurate diagnosis and preventing unnecessary radical treatments for presumed malignancies.

Asymptomatic and benign ectopic prostatic tissues typically do not require resection; however, they can be a source of PSA, and primary adenocarcinoma may occasionally develop from them (6, 16). Although benign ectopic prostate tissue does not inherently require excision, the potential for malignant transformation remains a significant concern. Adenocarcinoma can arise from ectopic prostate tissue, posing a risk if left untreated. The fact that this patient presented with a progressively enlarging mass warrants consideration of long-term surveillance strategies for similar cases, as ectopic prostate tissue may not remain dormant indefinitely. Additionally, ectopic prostate tissue can contribute to elevated PSA levels, particularly in post-prostatectomy patients, complicating oncologic follow-up.

Ectopic prostatic tissue is histologically and immunohistochemically indistinguishable from normal prostatic tissue. Radiological interpretation is challenging due to the rarity of such cases, and there are no characteristic findings on computed tomography scans. In our study, sarcoma and SCC were considered in the differential diagnosis of this perineal mass; however, it did not exhibit malignant characteristics and was well-defined.

Given the rarity of ectopic prostate tissue, imaging modalities such as pelvic MRI lack standardized criteria to differentiate benign ectopic prostatic tissue from malignant tumors. The solid-cystic nature of the perineal lesion initially raised suspicion of sarcoma or SCC, demonstrating how ectopic prostate tissue can mimic aggressive malignancies. This highlights an important gap in current diagnostic approaches, as MRI findings alone may not be sufficient to rule out malignancy, thereby necessitating biopsy or histopathological evaluation. The lack of typical imaging patterns for ectopic prostate tissue further complicates its identification, making clinical suspicion and interdisciplinary diagnostic strategies crucial for accurate assessment.

Our case is exceptionally unique due to its unusual perineal location, large mass formation, and the diagnostic dilemma it posed. While most reported cases of ectopic prostate tissue have involved microscopic or incidental findings, this case presented as a clinically significant lesion that required surgical resection due to its size, growth pattern, and imaging characteristics. Moreover, the potential for misdiagnosis as a malignant tumor emphasizes the importance of including ectopic prostate tissue in the differential diagnoses for unexplained perineal masses.

While this case provides valuable insights into the rare occurrence of ectopic prostate tissue, several limitations must be acknowledged. First, due to its rarity, the findings are based on a single case report, which limits the generalizability of conclusions regarding clinical behavior and management. Second, although MRI has proven effective for detection, the absence of standardized imaging criteria for ectopic prostate tissue presents challenges in differentiation it from malignancies. Third, the etiology of prostatic ectopia remains speculative, requiring further studies to confirm the underlying mechanisms. Lastly, long-term follow-up data on potential recurrence or malignant transformation are lacking, emphasizing the need for broader research and multi-institutional case reviews to better understand the clinical implications of this condition.

Conclusion

Ectopic prostate tissue, although rare, presents a unique diagnostic challenge due to its diverse locations and potential clinical implications. While it is typically asymptomatic, it can mimic malignancies, necessitating careful pathological and radiological evaluation to ensure an accurate diagnosis.

This case highlights the importance of considering ectopic prostate tissue in the differential diagnosis of perineal masses, especially in post-prostatectomy patients with elevated PSA levels. MRI remains a valuable tool for detection, and immunohistochemical analysis plays a critical role in confirming the diagnosis. Further research is required to better understand the etiology and potential malignant transformation of ectopic prostate tissue, which could enhance to improved patient management and outcomes.

Authors' Contribution

Concept and design: S.V.H. and M.M.S.; Acquisition,

analysis, and interpretation of data: M.M.S., A.A.F., and M.S.; Drafting of the manuscript: M.M.S.; Critical review of the manuscript for important intellectual content: S.V.H. and M.M.S.; Supervision: S.V.H., M.M.S., A.A.F., and M.S. All authors have reviewed the final version to be published and have agreed to be accountable for all aspects of the work.

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