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CASE REPORT

Intratesticular Leiomyoma Associated with Polyorchidism: A Rare Case Report

Poliorşidizm ile Birlikte Görülen İntratestiküler Leiomyom: Nadir Bir Olgu Sunumu

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ABSTRACT
Polyorchidism is an exceptionally rare congenital urogenital anomaly characterized by the presence of three or more testes, whereas testicular leiomyomas are uncommon benign tumors originating from smooth muscle cells and typically demonstrating slow growth. We report the case of a 74-year-old man with congenital polyorchidism who presented with suspected testicular malignancy. The patient was admitted with a complaint of firmness in the left scrotum. Physical examination revealed two normally palpable testes; however, a well-circumscribed, firm mass measuring approximately 3–4 cm in diameter was detected distal to the left inguinal canal. Serum tumor markers were within normal limits. Left inguinal exploration performed under spinal anesthesia demonstrated a normal left testis as well as a separate mass originating from the testicular base and terminating in an additional spermatic cord structure, which was subsequently excised. Histopathological examination confirmed the diagnosis of an intratesticular leiomyoma arising from a supernumerary testis containing a spermatic cord structure. Because intratesticular leiomyomas are rare and may clinically mimic malignant testicular tumors, definitive diagnosis relies solely on histopathological evaluation, often leading to radical orchiectomy in suspected cases. Polyorchidism has been reported in only a limited number of cases in the literature, and to the best of our knowledge, no tumor arising from a third testis has previously been described.

Keywords: Polyorchidism, testicular tumor, triorchidism, leiomyoma

ÖZET
Poliorşidizm, üç veya daha fazla testisin bulunmasıyla karakterize, oldukça nadir görülen konjenital bir ürogenital anomalidir; testiküler leiomyomlar ise düz kas hücrelerinden köken alan, yavaş büyüme eğilimi gösteren ve benign karakterde seyreden nadir tümörlerdir. Bu çalışmada, doğuştan üç testisi bulunan ve testiküler malignite şüphesiyle başvuran 74 yaşındaki bir erkek olgu sunulmaktadır. Sol skrotumda sertlik yakınmasıyla başvuran hastanın fizik muayenesinde iki testis normal olarak palpe edilirken, sol inguinal kanal distalinde yaklaşık 3–4 cm çapında, sınırlı ve sert bir kitle saptandı; tümör belirteçleri normal sınırlarda idi. Spinal anestezi altında gerçekleştirilen sol inguinal eksplorasyonda sol testisin normal olduğu, ayrıca testis tabanından kaynaklanan ve ikinci bir spermatik kord yapısı ile sonlanan ayrı bir kitlenin bulunduğu gözlendi ve lezyon eksize edildi. Histopatolojik incelemede spermatik kord yapısı içeren ikinci testise ait intratestiküler leiomyom tanısı konuldu. İntratestiküler leiomyomlar nadir görülmeleri ve klinik olarak malign testis tümörlerini taklit edebilmeleri nedeniyle kesin tanısı yalnızca histopatolojik değerlendirme ile konulabilen lezyonlar olup, bu şüphe nedeniyle çoğu olguda radikal orşiektomi uygulanmaktadır. Poliorşidizm ise literatürde son derece sınırlı sayıda bildirilmiş olup, üçüncü testisten köken alan tümör varlığına ilişkin bugüne kadar bildirilmiş bir olgu bulunmamaktadır.

Anahtar Kelimeler: Poliorşidizm, testis tümörü, üç testis, leiomyom

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INTRODUCTION

Polyorchidism is a rare congenital condition defined by the presence of more than two testicles. Since its first description in the late nineteenth century, fewer than a few hundred cases have been reported, highlighting its exceptional rarity (1). The embryological origin of polyorchidism has not been fully elucidated; however, several theories have been proposed. The most widely accepted hypothesis suggests abnormal division of the genital ridge during early embryogenesis, occurring before or during differentiation of the primordial gonads. Depending on the timing and extent of this division, the supernumerary testis may or may not have an associated epididymis or vas deferens. Alternative theories include transverse or longitudinal duplication of the genital ridge and incomplete degeneration of mesonephric tissue, all of which may contribute to the wide anatomical variability observed in reported cases.The most frequent form is triorchidism, in which the supernumerary testis is usually located

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within the scrotum. From an anatomical and functional standpoint, polyorchidism demonstrates considerable heterogeneity. The supernumerary testis may be located within the scrotum, inguinal canal, or abdomen, with scrotal localization being the most common. Classification systems have been proposed based on reproductive potential and anatomical connections to the epididymis and vas deferens, as these features have implications for fertility preservation and management strategies. Testicular positioning is reported to be scrotal in 75% of cases, inguinal in 20%, and abdominal in 5% (2,3). Supernumerary testes may be unilateral or bilateral, with over half of reported cases involving the left side (4).

While the majority of testicular tumors are malignant, benign masses may also occur. These include epidermoid cysts, lipomas, benign teratomas, and sex cord–stromal tumors (5). Testicular leiomyomas are particularly rare and arise from smooth muscle proliferation, typically exhibiting slow growth (6,7). In this report, we present a case of triorchidism with leiomyoma arising from the third testis following complaints of scrotal firmness.

CASE

A 74-year-old male patient presented to our clinic with complaints of firmness in the left scrotum. On examination, both testicles were normal in size and consistency. However, a firm, well-circumscribed mass measuring approximately 3–4 cm was palpated in the distal left inguinal canal at the level of the spermatic cord. Scrotal ultrasonography confirmed the presence of a 3 cm well-defined lesion in the same region. Tumor markers were as follows: Lactate Dehydrogenase (LDH): 186 U/L (normal: 125–220 U/L), Alpha-fetoprotein (AFP): 1.34 ng/mL (normal: 0–10 ng/mL), and β-Human Chorionic Gonadotropin (β-HCG): <0.2 U/L (normal: 0–2 U/L).

Based on these findings, surgical exploration was planned. Under spinal anesthesia, a left inguinal incision was performed. The testicle and the mass were exposed, revealing two separate spermatic cord structures. The left testicle appeared normal, while the second cord terminated in a mass arising from the base of an accessory testis (Figure 1). The mass was completely excised. The patient was discharged on the first postoperative day without complications. A follow-up scrotal ultrasound showed no additional pathology, and the patient remained asymptomatic at the 6-month postoperative follow-up.

Histopathological evaluation revealed a 3 × 2.5 × 2.5 cm well-circumscribed mass with a smooth external surface and an associated 3 × 0.8 cm segment of spermatic duct. The lesion consisted of spindle-shaped smooth muscle cells with focal nuclear atypia but no mitotic figures or necrosis, consistent with leiomyoma (Figures 2–3). Written informed consent was obtained from the patient prior to surgery.

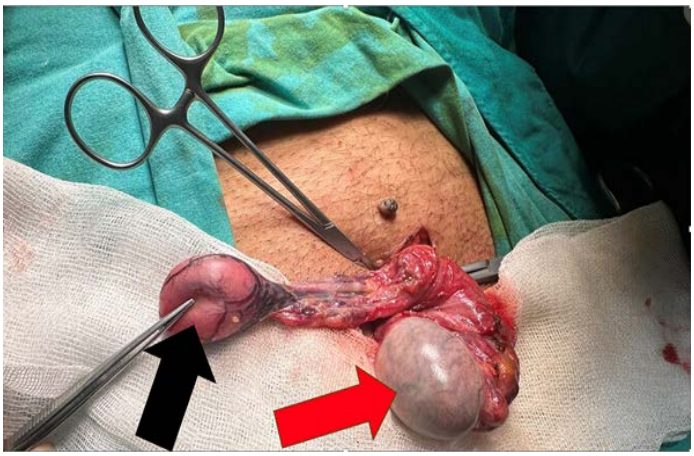

Figure 1. Macroscopic image of the mass detected after a left inguinal incision during surgery and the left testicle. Red arrow: left testicle, Black arrow: leiomyoma of the 3rd testicle.

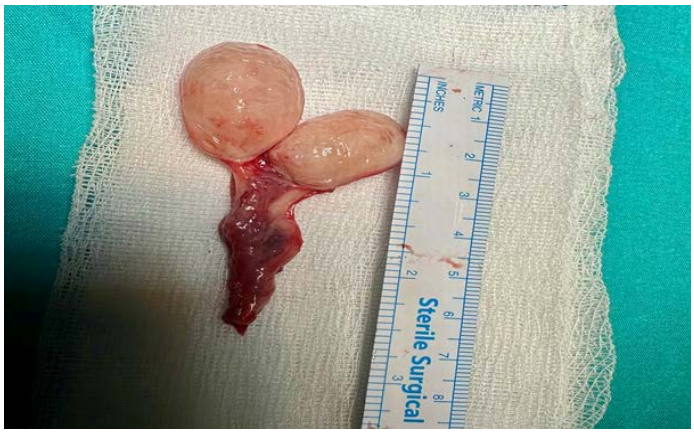

Figure 2. Postoperative macroscopic image of a specimen taken after mass excision. The mass is approximately 3 cm in size and appears to have smooth borders.

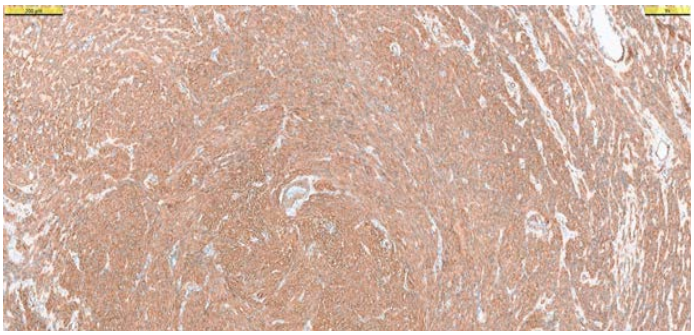

Figure 3. Microscopic examination reveals a well-defined, encapsulated mass of smooth muscle tissue organized into interwoven bundles. The cells have long, spindle-shaped nuclei with rounded ends, heterogeneous chromatin, thin nucleoli, and eosinophilic cytoplasm with indistinct cytoplasmic boundaries. Extensive staining with smooth muscle actin (SMA) stain is observed.

DISCUSSION

Leiomyomas are benign tumors originating from smooth muscle cells. Although they are most commonly found in the renal pelvis, they may also arise in the bladder, spermatic cord, epididymis, prostate, glans penis, or scrotal structures (8). These tumors are generally observed in individuals older than 50 years. Previous reports have described leiomyomas in locations such as the tunica vaginalis, tunica albuginea, epididymis, spermatic cord, and even the testicular parenchyma (9). Scrotal leiomyomas typically present as slow-growing masses. Due to their indolent course, the interval between initial detection and hospital admission can range from 2 to 20 years (10). On physical examination, they usually appear as firm, non-tender nodules, often localized to the pole of the testis. Their mean size is approximately 3 cm, although reported sizes range from 1 to 8 cm (11). A wide variety of differential diagnoses should be considered, including fibroma, supernumerary testis, and sebaceous cyst (12). Painful schwannomas associated with scrotal ulcerations may clinically mimic squamous cell carcinoma, particularly when preceded by trauma (13). Tumor markers are usually within normal limits, as observed in our patient.

Pathological evaluation of leiomyomas relies on several criteria, including tumor size greater than 5 cm, irregular margins, more than 10 mitotic figures per 10 high-power fields, and significant cytological atypia. A lesion demonstrating one of these features may be classified as a leiomyoma; two features suggest an atypical leiomyoma, while three or more are indicative of leiomyosarcoma (14). In the present case, the mass was well-demarcated, measured less than 5 cm, and showed no mitotic activity or necrosis, supporting the diagnosis of benign leiomyoma. From a management perspective, a recent systematic review of epididymal leiomyomas demonstrated that testis-sparing surgical approaches are both feasible and safe, with reported procedures ranging from simple lesion excision to epididymectomy. Importantly, no recurrences were documented during follow-up. These findings support organ-preserving strategies when tumor markers are negative, imaging does not suggest invasive disease, and intraoperative findings are consistent with a benign, well-circumscribed paratesticular lesion (15).

Polyorchidism is an uncommon congenital condition, most frequently involving a supernumerary testis on the left side (16). Rare cases of bilateral or triplicated testes have also been reported. Although its etiology remains speculative, polyorchidism is generally thought to result from abnormal division of the genital ridge, with or without involvement of the Wolffian duct (17). A supernumerary testis may mimic other scrotal pathologies, including tumors, spermatoceles, varicoceles, or hydroceles, and may also be incidentally detected during surgery for undescended testes (18). Therefore, physical examination alone is insufficient for diagnosis. Imaging modalities—particularly Doppler ultrasonography supplemented by magnetic resonance imaging (MRI)—play a crucial role in evaluation (19). A recent systematic review reported that imaging-based diagnosis was common, and while observation was frequently chosen, surgical intervention was preferred when the supernumerary testis was ectopic or when features raised concern for neoplastic transformation (20). In the present case, the third testis was identified intraoperatively by the presence of two distinct spermatic cords and ductus deferens structures. Bergholz et al. proposed a classification system for polyorchidism (16):

- Type A: Testis with drainage via the vas deferens
  - A1: Own epididymis and vas deferens
  - A2: Own epididymis but shared vas deferens
  - A3: Shared epididymis and duct
- Type B: Testis without drainage
  - B1: With epididymis
  - B2: Without epididymis; isolated testicular tissue

Our case is consistent with Type A1 polyorchidism, as the supernumerary testis possessed its own ductal structures. The optimal management of polyorchidism remains controversial. The balance between the potential risk of malignancy and the reproductive contribution of the supernumerary testis presents a clinical dilemma. While most authors advocate preservation of the supernumerary testis, orchiectomy is recommended when malignancy is suspected (16). The coexistence of polyorchidism and leiomyoma is particularly challenging, as polyorchidism itself may present as an "additional mass," and the presence of a solid paratesticular lesion may further increase suspicion of malignancy, potentially leading to overtreatment. This is especially relevant because imaging findings may not reliably distinguish benign from malignant paratesticular tumors. Recent imaging-focused studies emphasize that accurate preoperative identification of benign extratesticular lesions is essential to avoid unnecessary orchiectomy, and that MRI can be valuable in clarifying lesion origin and anatomical relationships (21).

The unique contribution of this report lies in highlighting a rare but clinically relevant scenario in which a congenital anatomical variant (polyorchidism) coexists with a benign paratesticular smooth muscle tumor (leiomyoma). This combination increases diagnostic uncertainty and the risk of unnecessary radical surgery. Our case underscores the importance of differentiating diagnostic pitfalls related to polyorchidism—such as misinterpretation of accessory testicular tissue—from those related to leiomyoma, which may closely mimic malignant disease. In carefully selected patients with reassuring tumor markers and non-aggressive imaging findings, a personalized, testis-sparing surgical approach with definitive histopathological confirmation can achieve oncological safety while minimizing morbidity.

CONCLUSION

Polyorchidism is a rare congenital anomaly that can complicate the evaluation of scrotal masses, particularly when associated with paratesticular tumors. This case illustrates the uncommon coexistence of a supernumerary testis and a paratesticular leiomyoma, a benign entity that may closely mimic testicular malignancy. Given the limitations of imaging in such rare anatomical settings, careful clinical judgment

is essential. In selected patients with a low suspicion of malignancy, testis-sparing surgery represents a reasonable and effective approach, with definitive diagnosis established by histopathological examination.

DECLARATIONS

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