

## CASE REPORT

# Polyorchidism: radiological features and therapeutic management of a rare congenital anomaly

Iwona Anderko<sup>1</sup>, Paweł Juszcak<sup>2</sup>, Przemysław Mańkowski<sup>2</sup>, Patrycja Sosnowska-Sienkiewicz<sup>3</sup>

<sup>1</sup>Student Scientific Society, Poznan University of Medical Sciences, Poznan, Poland

<sup>2</sup>Department of Pediatric Surgery, Traumatology and Urology, Poznan University of Medical Sciences, Poznan, Poland

<sup>3</sup>Department of Pediatric Surgery, Medical University of Warsaw, Warsaw, Poland

## ABSTRACT

Polyorchidism, or the presence of supernumerary testes, is a rare congenital anomaly of the genitourinary system. Most cases are asymptomatic and incidentally discovered during imaging or surgery for unrelated conditions. The anomaly is associated with an increased risk of torsion, inguinal hernia, cryptorchidism, and, rarely, malignancy. Accurate diagnosis requires careful evaluation of anatomical and functional characteristics using high-resolution ultrasonography, magnetic resonance imaging, and histopathological assessment when indicated. Management should be individualized, balancing fertility preservation with malignancy risk, and may involve either conservative monitoring or surgical intervention. Psychological and ethical considerations are important, particularly for young patients, highlighting the need for multidisciplinary care involving urologists, pediatric surgeons, pediatricians, and psychiatrists. We present a rare case of left-sided triorchidism in a 17-year-old male, emphasizing the diagnostic challenges and the importance of an integrated, patient-centered approach in both medical and psychosocial aspects of care.

## KEY WORDS:

pediatric surgery, polyorchidism, supernumerary testis, triorchidism, genitourinary abnormalities.

## INTRODUCTION

Polyorchidism, commonly referred to as supernumerary testis, is an uncommon congenital defect of the genitourinary system which is characterized by having more than two testes. This exceedingly rare condition has been documented in fewer than 250 cases in medical literature. The estimated occurrence of this anomaly is approximately 1 in 1,000,000 males [1–3]. The first recorded case appeared in the literature through Blasius' observations during a routine autopsy in 1670, while the earliest histologically validated diagnosis was made by Lane in 1895 [4]. The exact cause of polyorchidism remains unclear. The most widely recognized theory posits that an unintentional division of the genital ridge occurs before the eighth week of embryonic development. The location at which this trans-

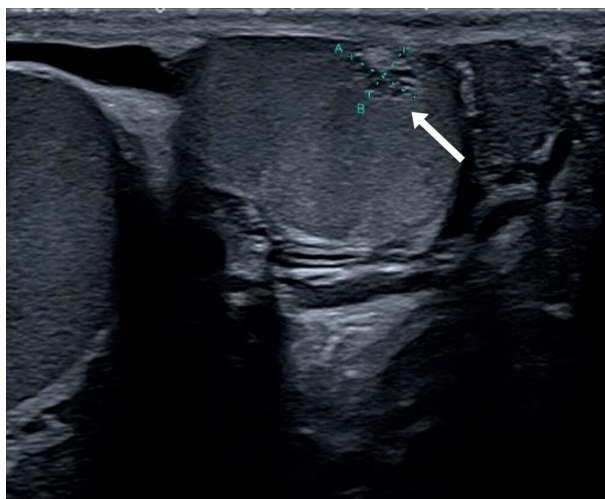
verse split happens influences the anatomical arrangement of the supernumerary testis, including whether there is an additional vas deferens or epididymis present [5]. Other embryological theories have been suggested, such as irregular cell migration within the genital ridge, duplication or partial splitting of the urogenital ridge, incomplete regression of mesonephric structures, or the creation of peritoneal bands [3]. Most cases remain asymptomatic and are discovered by chance, often during the evaluation of a scrotal or inguinal mass. Approximately half of the patients are diagnosed between the ages of 15 and 25 [6].

## CASE REPORT

A 17-year-old male patient was admitted to the pediatric surgery department for evaluation of a suspected

## ADDRESS FOR CORRESPONDENCE:

Iwona Anderko, Poznan University of Medical Sciences, 10 Fredry St., Poznan, Poland,  
e-mail: iwonaanderko00@gmail.com

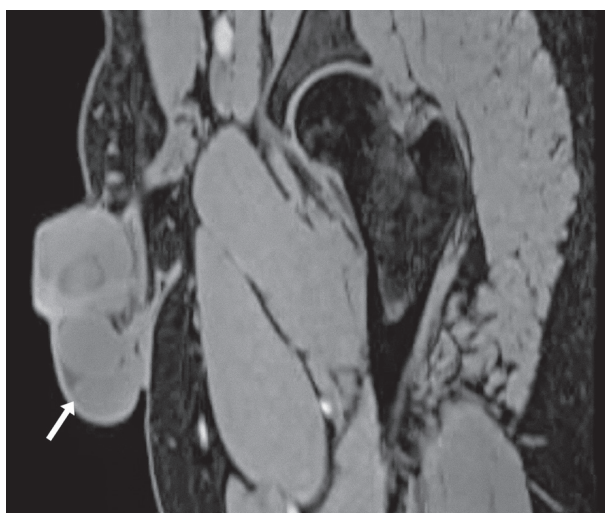


**FIGURE 1.** Scrotal ultrasound of the supernumerary left testis ( $22 \times 21 \times 17$  mm) with a  $5.5 \times 4$  mm heterogeneous area in inferior portion (white arrow)

*Vascular flow matches surrounding tissue. The finding is indeterminate.*



**FIGURE 2.** Coronal T2-weighted magnetic resonance imaging showing the supernumerary left testis ( $22 \times 20 \times 16$  mm) with a  $6 \times 4$  mm hyperintense focal lesion suggestive of the cystic process (white arrow), although malignancy cannot be excluded



**FIGURE 3.** Sagittal T2 magnetic resonance imaging showing the supernumerary left testis ( $22 \times 20 \times 16$  mm) with a  $6 \times 4$  mm hyperintense focal lesion suggestive of the cystic process (white arrow), although malignancy cannot be excluded

congenital testicular anomaly after an abnormality in the left hemiscrotum was palpated during a urological consultation for acute right testicular pain following a minor trauma. He reported no previous scrotal symptoms, and both his personal and family histories were unremarkable for genitourinary disorders. Scrotal ultrasonography demonstrated marked asymmetry and the presence of two distinct testes within the left hemiscrotum. The lower testis measured  $22 \times 21 \times 17$  mm and contained, in its inferior portion, a well-defined heterogeneous area measuring approximately  $5.5 \times 4$  mm, with vascular flow comparable to the surrounding testicular tissue (Figure 1). As the ultrasound findings were inconclusive, magnetic resonance imaging (MRI) was performed. Magnetic resonance imaging confirmed the pres-

ence of a supernumerary testis in the lower part of the left hemiscrotum, measuring  $22 \times 20 \times 16$  mm, with a small peripheral lesion approximately  $6 \times 4$  mm in size. The orthotopic left testis measured  $33 \times 28 \times 23$  mm and the right testis  $46 \times 28 \times 26$  mm, both showing normal morphology. Small bilateral hydroceles were noted, and no abnormalities of the epididymides, spermatic cords, or inguinal lymph nodes were observed (Figures 2, 3). Preoperative tumor markers, including  $\alpha$ -fetoprotein,  $\beta$ -human chorionic gonadotropin, and lactate dehydrogenase, were within normal limits. Given the presence of a focal lesion within the supernumerary testis and the indeterminate imaging findings, the case was discussed at a multidisciplinary tumor board involving pediatric surgeons, radiologists, and pediatric oncologists. Based on the imaging characteristics and the concerns raised, particularly by radiologists and oncologists, the decision was made to proceed with surgical removal of the supernumerary testis containing the MRI-detected abnormality. A transverse skin incision was made above the left inguinal canal, followed by dissection through the aponeurosis of the external oblique muscle to expose the canal. The spermatic cord, including the cremaster muscle, was carefully isolated, and both the orthotopic and supernumerary testes were mobilized within their tunicae. Intraoperative inspection revealed that the testes shared a common epididymis and vas deferens (Bergholz type A3, Leung type 2). The supernumerary testis was smaller, firm in consistency, and contained a visible pathological lesion. Using meticulous sharp and blunt dissection, the additional gonad was excised without any injury to the epididymis or vas deferens. The orthotopic testis appeared normal and was repositioned into the scrotum (Figure 4). Hemostasis was achieved, the inguinal canal was reconstructed, and the skin was closed with vertical mattress sutures. The procedure was com-

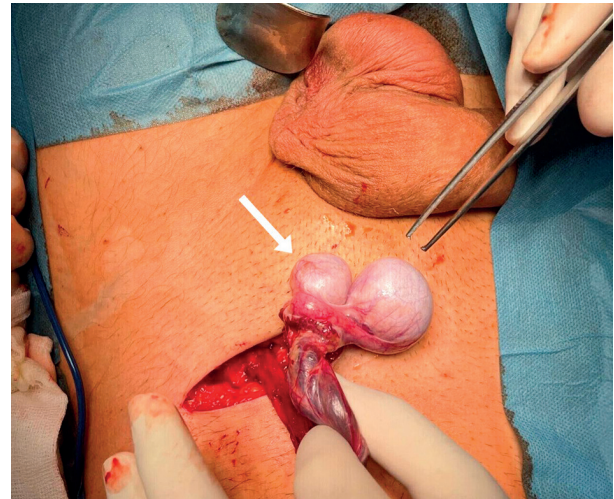
pleted without intraoperative complications. The excised specimen measured  $2.6 \times 2.1 \times 1.8$  cm and was submitted for histopathological examination. Histology revealed a testis with preserved architecture and active spermatogenesis, accompanied by focal edema and mild chronic inflammatory infiltrates. No malignant features were identified. The postoperative course was uneventful, and the patient was discharged in good general condition. At follow-up, the scrotum appeared symmetrical with a satisfactory cosmetic outcome, and the patient reported no discomfort, swelling, or aesthetic concerns.

## DISCUSSION

Triorchidism, which is defined as the presence of three testes, is the most common form of polyorchidism. In most of the cases that have been documented, the additional testis is found on the left side of the scrotum (approximately 65%) [4, 7]. In terms of anatomical distribution, supernumerary testes are most often found in the scrotal sac (around 76%), followed by the inguinal canal (23–24%), and less commonly in the abdominal cavity (around 9%) [5]. The typical age at diagnosis is around 17 years old [3, 7]. Similarly, in our case, the condition was diagnosed at the age of 17, with the supernumerary testis located in the left hemiscrotum.

Most individuals with polyorchidism are asymptomatic and are found unintentionally during imaging or surgery for unrelated urogenital issues. However, it may coexist with other abnormalities, such as an inguinal hernia (15–30%), cryptorchidism (15–30%), testicular torsion (9–15%), a hydrocele (9%), and more rarely, malignancy (reported in fewer than 6% of cases) [3, 7]. In our case, symptoms of pain and swelling originated from the normally developed right testis, while the left side presented only with asymmetry due to the presence of a supernumerary testis.

Several classification systems have been proposed for describing polyorchidism. The most widely accepted system is that of Bergholz *et al.* [8], which categorizes cases based on the presence and configuration of drainage through the vas deferens. According to this classification, polyorchidism is divided into two main types based on whether the additional testis has a drainage system through the vas deferens. Type A includes supernumerary testes that are connected to the vas deferens, and it has three subtypes. In type A1, the extra testis has its own epididymis and vas deferens. In type A2, it has its own epididymis but shares a vas deferens with the normal testis. Type A3, which is the most common, describes a situation where the extra testis shares both the epididymis and the vas deferens with the neighboring testis. Type B refers to supernumerary testes that are not connected to the vas deferens. It includes type B1, in which the testis has an epididymis but no vas deferens, and type B2, which consists only of testicular tissue without any ducts or excretory structures [2, 5]. Another classification pro-



**FIGURE 4.** Intraoperative photo showing left triorchidism

The supernumerary testis (white arrow) is attached to a shared spermatic cord and epididymal complex, appearing identical in size and texture to the orthotopic testis.

posed by Leung is based on embryological development. It distinguishes four forms:

Type 1 – the extra testis has no epididymis or vas deferens,

Type 2 – it shares both structures with the normal testis on the same side,

Type 3 – it has its own epididymis but shares a vas deferens,

Type 4 – representing a complete duplication of the testis, epididymis, and vas deferens [2, 5] (Table 1).

In our patient, the anomaly was classified as Bergholz *et al.* [8] type A3 and Leung type 2, meaning that the extra testis shared both the epididymis and the vas deferens with the neighboring testis, which could possibly affect its function.

One of the most critical clinical concerns associated with polyorchidism is the significantly increased risk of malignant transformation in supernumerary testes. Reported studies estimate the overall risk of testicular cancer in supernumerary testes at 5.7–6.4%, which is higher than in the general male population (0.004%) and in isolated undescended (cryptorchid) testes without supernumerary tissue (0.045%) [3, 6, 7, 9]. However, it is important to emphasize that approximately 88% of testicular cancers in polyorchidism occur in supernumerary testes located outside the scrotum, usually in inguinal or abdominal positions [10]. Therefore, there is currently no clear evidence that an accessory testis located within the scrotum carries a significantly higher risk of malignancy compared with normally descended testes. The most reported histological types of tumors in these cases include seminomas, teratomas, and choriocarcinomas [6, 10]. In our patient, histology showed preserved testicular structure with active spermatogenesis and no signs of malignancy.

Imaging plays a key role in both the diagnosis and management of polyorchidism. High-resolution ultrasonography is typically the initial modality, as it can iden-

TABLE 1. Comparison of polyorchidism classification systems

| Classification system                                    | Type | Description                                                                 | Epididymis | Vas deferens |
|----------------------------------------------------------|------|-----------------------------------------------------------------------------|------------|--------------|
| Bergholz <i>et al.</i><br>(anatomical<br>classification) | A1   | Extra testis with its own epididymis and vas deferens                       | Separate   | Separate     |
|                                                          | A2   | Own epididymis, shared vas deferens with normal testis                      | Separate   | Shared       |
|                                                          | A3   | Shares both epididymis and vas deferens with normal testis<br>(most common) | Shared     | Shared       |
|                                                          | B1   | Has epididymis but no vas deferens (no drainage)                            | Present    | Absent       |
|                                                          | B2   | Only testicular tissue, no ducts                                            | Absent     | Absent       |
| Leung<br>(embryological<br>classification)               | 1    | No epididymis or vas deferens                                               | Absent     | Absent       |
|                                                          | 2    | Shares both with normal testis on the same side                             | Shared     | Shared       |
|                                                          | 3    | Own epididymis, shared vas deferens                                         | Separate   | Shared       |
|                                                          | 4    | Complete duplication of the testis, epididymis, and vas deferens            | Separate   | Separate     |

tify a supernumerary testis by its similar echotexture and vascularity compared to the adjacent normal testis [5]. When ultrasound findings are unclear or further anatomical detail is required, MRI provides valuable confirmation. On MRI, additional testes show signal characteristics on T1- and T2-weighted images that closely resemble normal testicular tissue [10–12]. Advanced imaging techniques such as contrast-enhanced ultrasound can also aid in evaluating vascular patterns, helping to distinguish benign from malignant lesions and potentially reducing the need for unnecessary surgical intervention [5, 13]. In our patient, MRI and ultrasound detected a supernumerary testis within the left hemiscrotum, including a small lesion that could not be ruled out as potentially malignant. Surgical removal was therefore performed because of this indeterminate finding, as assessed by a multidisciplinary team. Histopathology ultimately confirmed the diagnosis and revealed no malignancy.

Due to its rarity, there is no universally accepted approach to managing polyorchidism, and treatment decisions are guided by factors such as the anatomical location of the supernumerary testis, patient age, reproductive potential, and suspicion of malignancy [9, 14, 15]. Preserving fertility is an important consideration, as studies indicate that approximately 50–65% of supernumerary testes maintain active spermatogenesis [16, 17]. When the additional testis is located within the scrotum, appears normal, and has intact vascular supply and drainage, a conservative approach is generally preferred. This includes regular clinical follow-up, patient self-examinations, and imaging surveillance using ultrasound or MRI [14, 18, 19]. Testes located outside the scrotum or showing suspicious features on imaging may require surgical removal due to a higher malignancy risk, estimated at 5–6%, primarily in undescended supernumerary testes [14, 20]. Classification systems, such as that proposed by Bergholz *et al.* [8], which differentiate supernumerary testes based on the presence of drainage pathways, help guide management: testes with drainage (type A)

are more likely to be preserved, whereas those without (type B) have limited reproductive potential and higher malignancy risk [5, 9]. In cases of ectopic supernumerary testes, particularly in younger patients, orchidopexy is recommended to preserve fertility and reduce the risk of torsion. In older patients or when malignancy is suspected, orchiectomy is indicated [5, 9]. Emerging fertility preservation techniques, including cryopreservation of germ cells or testicular tissue, may offer future options but remain largely experimental [21]. In our case, histological examination confirmed active spermatogenesis in the additional testis, suggesting fertility could have been preserved; however, surgical excision was performed due to a suspicious lesion to establish a definitive diagnosis.

Long-term monitoring and follow-up in polyorchidism present difficulties due to the condition's rarity and the absence of standardized management protocols. Most recommendations suggest an ultrasound every 6–12 months to observe any morphological changes in the testes or to detect early signs of malignancy. Additional imaging, such as MRI, may be indicated when ultrasound findings are inconclusive or when there is a suspicion of neoplastic changes [2, 5, 13, 14]. Routine laboratory testing, including tumor marker evaluation, is generally reserved for cases where clinical or imaging findings raise concern for malignancy, rather than applied universally to all patients. Several studies indicate that an active surveillance strategy incorporating regular clinical assessments, patient self-examination, and imaging can be a safe and effective approach for individuals managed conservatively, particularly when the supernumerary testis is in the scrotum and shows no evidence of neoplastic transformation [5, 12]. Due to the lack of formal guidelines, follow-up plans should be tailored to each patient, considering factors such as age, associated anomalies, anatomical location of the supernumerary testis, and adherence to clinical monitoring. This individualized approach allows for effective cancer surveillance while minimizing unnecessary interventions [2, 5, 12].

## CONCLUSIONS

Polyorchidism is a rare condition that presents unique diagnostic and therapeutic challenges. Careful evaluation of the anatomical and functional characteristics of supernumerary testes is essential to guide management and preserve fertility. This evaluation should primarily rely on detailed imaging, while histopathological assessment may be considered in selected cases where imaging or clinical findings raise concern. For supernumerary testes within the scrotum that show no concerning features, the recommended management is careful observation, including regular imaging and routine self-examination. Long-term follow-up should be individualized, including regular imaging and self-monitoring, to detect any potential early signs of malignancy. Optimal medical outcomes require multidisciplinary care, including a pediatric urologist, a pediatric surgeon, and pediatricians. This case underscores the importance of a comprehensive, integrated approach to patient care.

## DISCLOSURES

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2. Assistance with the article: None.
3. Financial support and sponsorship: None.
4. Conflicts of interest: None.

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