International Surgery Journal
Valente T et al. Int Surg J. 2022 Dec;9(12):2059-2061
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20223173

Case Report

A rare case of polyorchidism in a 48-year-old man with inguinal hernia

Tânia Valente[1]*, Nuno Ferreira[1], Gonçalo Ferreira[1], Arnaldo Figueiredo[1], Patrícia Bárbara[1], Cristina Aniceto[1], Ricardo Marinho[1], Miguel C. Santos[1], Pedro Eufrásio[2]

[1]Department of Surgery, [2]Department of Urology, Centro Hospitalar de Leiria, Leiria, Portugal

Received: 05 October 2022
Accepted: 04 November 2022

***Correspondence:**
Dr. Tânia Valente,
E-mail: tania.telmo.valente@gmail.com

ABSTRACT

Polyorchidism is a rare congenital anomaly characterized by the presence of more than two testes. Triorchidism is the most common variant. Polyorchidism has been reported in around two hundred cases in the world to date. It is usually diagnosed incidentally in routine exams or surgery. It can be associated with undescended testis (40%), hernia (30%), and torsion (15%), among others. Given how rare it is, polyorchidism requires a high degree of suspicion. The exact aetiology of polyorchidism is unknown. The preservation of supernumerary testis has been controversial. Today, in the absence of pain, conservative management with a strict ultrasound and magnetic resonance imaging (MRI) follow-up is the most widely used approach. We present a case of a 48-year-old man who had a personal history of left cryptorchidism and came to our appointment with a bilateral inguinal hernia. During surgery for hernia repair, we found a supernumerary testis located on the left inguinal region with no epididymis or vas deferens. We opted for orchidopexy. In the other left testis, we did an orchidectomy due to the presence of a suspicious nodule. The histology was negative for neoplasia.

Keywords: Polyorchidism, Supernumerary testis, Orchidectomy, Orchidopexy

INTRODUCTION

Polyorchidism is a rare congenital anomaly characterized by the presence of more than two testes. Triorchidism (presence of three testes) is the most common variant although there have been described cases of males with four or even five testes. The first case of triorchidism was reported by Blasius in 1670 at a routine autopsy.[1] The first histologically proven triorchidism was reported by Lane in 1895.[1,8]

Polyorchidism has been reported in around two hundred cases in the world to date.

The exact aetiology of polyorchidism is unknown, but the proposed mechanism is the longitudinal or transverse division of the genital ridge possibly by the development of peritoneal bands during the 6th to 8th weeks of embryogenesis.

It is usually diagnosed incidentally in young asymptomatic patients in routine exams or surgical exploration.

This condition is associated with undescended testis (40%), hernia (30%), torsion (15%), hydrocele (9%), and malignancy (6%).[1,6] This last condition may include teratoma, anaplastic seminoma, choriocarcinoma, and rhabdomyosarcoma.[5]

Given how rare it is, polyorchidism requires a high degree of suspicion.

There are two important classifications of polyorchidism, the embryological one of Leung and the vascular of Berglozs et al.[6,7,11]

The treatment of polyorchidism is controversial and usually depends on the location of the testis, age, size of the testicle, and reproductive potential.[3]

We present a case of a 48-year-old man who had a personal history of left cryptorchidism and came to our appointment with a bilateral inguinal hernia. During surgery for hernia repair, we found a supernumerary testis located on the left inguinal region.

CASE REPORT

We present a case of a 48-year-old man who had a personal history of left cryptorchidism that was never repaired and came to our appointment with a bilateral inguinal hernia. He was healthy and had two children. On detailed physical examination, a reducible swelling in each inguinal region was noted with positive Valsalva's manoeuvre and positive deep ring occlusion test. Inguinal ultrasound confirmed bilateral hernia and revealed exteriorization of a hyporeflective nodular formation on the left inguinal region.

We proposed a transabdominal preperitoneal patch technique (TAPP) for repairing his inguinal hernias and the patient agreed.

During surgery for hernia repair, a mass on the left inguinal region was noted and thus we converted to an open anterior approach. We found a supernumerary testis located on the left inguinal region with no epididymis or vas deferens. We called for a urologist and together we opted for orchidopexy of the supernumerary testis. In the other left testis, we performed an orchidectomy because it contained a suspicious nodule. We proceeded with the TAPP approach to conclude the surgery.

There were no postoperative complications, and the patient was discharged on the first day. The histology of the testis with the suspicious nodule was negative for neoplasia.

He had no complaints or evidence of recurrence at 1-month, 6-month, and 1-year follow-up appointments. The pain disappeared after a few days and his sexual performance was not affected.

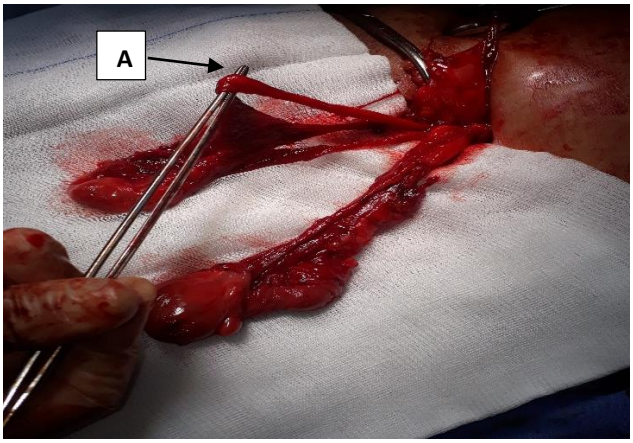

Figure 1: Intra-operative findings after left inguinal incision – two testicles. In (A) we can see the vas deferens.

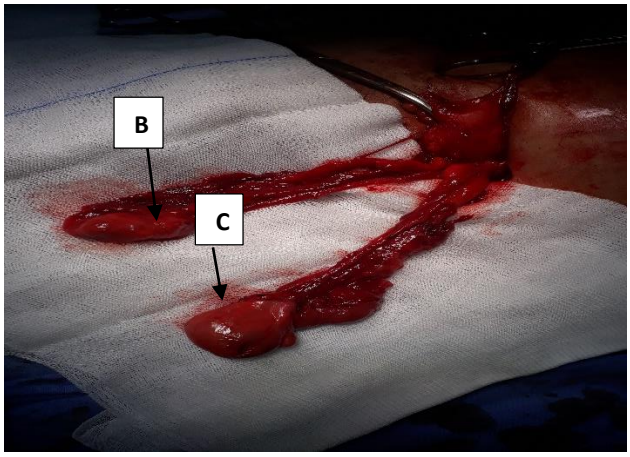

Figure 2: The suspicious nodule can be seen in the "normal testis" (B). The (C) corresponds to the supernumerary (SNT) testis, which has no deferens or epididymis.

DISCUSSION

Polyorchidism is a rare condition, with only around 200 cases reported in the literature. The supernumerary testis much more common on the left side and triorchidism is the most common condition, which was also the case of our patient. The exact etiology is unknown, as said before.

There are, in fact, three classifications for this condition. One is based on the location of the supernumerary testis - scrotal (66%), inguinal (23%), and abdominal (9%). In our case, the supernumerary test was located on the left inguinal region, as well as the other left testis.

In 1998, Leung described embryological development with functional implications as follows: type 1: supernumerary testis (SNT) without epididymis and vas deference; type 2: SNT shares common epididymis and vas deferens with ipsilateral testis; type 3: SNT has its own epididymis but shares a common vas deferens; and type 4: complete duplication of testis, epididymis, and vas.

Type 2 is the most common type and together with type 3 represents more than 90% of cases. Our case is even rarest corresponding to type 1.

Bergholz et al 2009 present an innovative classification based on the drainage system of the SNT testis. This one includes type A - testis being drained by an outflow path – and type B - undrained testes without connection to a draining vas deferens.

Type A testes are further divided into the subgroups A1 (own epididymis and vas deferens), A2 (own epididymis but common vas deferens), A3 (common epididymis but common vas deferens), A4 (own vas deferens but common epididymis) and AX (no further description of appendixes). Type B can also be divided into B1 (own epididymis), B2 (no epididymis, thus testicular tissue

only), and BX (no further description of appendixes). Our patient condition corresponds to a B2.

As said before, polyorchidism is mostly diagnosed incidentally during routine exams or during surgical exploration for groin surgery, with undescended testis comprising around 40%. In our patient, the condition was incidentally found during TAPP repair for bilateral inguinal hernia. Notice he had left cryptorchidism that was never corrected.

The differential diagnosis for this condition is spermatocele, encysted hydrocele, varicocele, aberrant epididymis, Morgagnian cyst, or testicular neoplasm.[4,5]

Polyorchidism can be diagnosed by ultrasound, which is the most used exam, doppler, or MRI.[1,4,5] Ultrasound can detect around 81% of cases but did not detect polyorchidism in our patient although it mentioned exteriorization of a hyporeflective nodular formation on the left inguinal region.[1] The preservation of supernumerary testis has been a controversial issue of discussion.

In the past, surgical removal of a supernumerary testicle, especially in an abdominal location, was performed due to the risk of malignancy. In scrotal and inguinal locations, some authors used to advocate the preservation of SNT testis with straightforward follow-up.

Today, the quality of imaging study changed the way we treat polyorchidism. In absence of pain (like our patient) we may choose conservative management a straight ultrasound and an MRI follow-up.

In our case, we removed the left testicle with a suspicious nodule and opted to do the orchidopexy in the supernumerary testis to preserve one testis on the left side. Macroscopically the SNT was normal and with orchidopexy, it is now located in the scrotum which becomes easier to recognize and diagnose a possible neoplasm in the future. In addition, the preservation of testis with an orchidopexy helps keep man's health by maintaining his endocrine function. The histology of the removed testicle was negative for neoplasia and thus we opted for surveillance.

CONCLUSION

Polyorchidism is a rare condition occurring mostly in young males and is discovered incidentally during routine exams or surgery. Despite his rarity, it should be considered as one of the differential diagnoses during groin surgery and in scrotal masses as well. The treatment of polyorchidism is still controversial and usually depends on the location of the testis, age, size of the testicle, and reproductive functionality.[3]

Today, the presence of imaging study changed the way we treat polyorchidism. In absence of pain, we may consider conservative management with a straight ultrasound and MRI follow-up.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Assefa HG, Erge MG, Gebreselassie HA. Triorchidism, a Rare Genitourinary Anomaly: A Case Series. Res Rep Urol. 2021;13:549-52.
2. Roy J, Hunt J, Ecclestone H, Johnston J. Three's a crowd: A very rare form of polyorchidism with sertoli cell adenomas. Urol Case Rep. 2020;34:101512.
3. Assefa HG, Sedeta AM, Gebreselassie HA. Polyorchidism during orchidopexy; A case report with review of literature. Urol Case Rep. 2021;39:101750.
4. Mazketly M, Aleter O, Brimo Alsaman MZ, Bazkke B, Jouda ME, Kayyali A. A rare case of polyorchidism in a 40-year-old man. A case report. Ann Med Surg (Lond). 2021;66:102389.
5. Núñez García B, Álvarez García N, Pérez-Gaspar M, Esteva Miró C, Santiago Martínez S, Betancourth Alvarenga JE, San Vicente Vela B, Jiménez Gómez J, Jiménez Arribas P. Polyorchidism in pediatric patients: a case report and a literature review. Cir Pediatr. 2021;34(3):160-3.
6. Bayissa BB, Tesfaye D. Triorchidism; an incidental finding at inguinal hernia repair: A case report. Int J Surg Case Rep. 2020;77:813-5.
7. Ojaghzadeh D, Mahmoudpour M, Ezzati N, Akhavi Milani A. Polyorchidism in ultrasound examination: A case report. Andrologia. 2021;53(1):e13832.
8. Mandalia U, Pakdemirli E. A case of triorchidism. Radiol Case Rep. 2020;15(9):1643-5.
9. Mittal PG, Peters NJ, Malik MA, Samujh R. Intraoperative Dilemmas in Polyorchidism: To Pex or not to Pex!! J Indian Assoc Pediatr Surg. 2020;25(3):175-7.
10. Boussaffa H, Naouar S, Ati N, Amri M, Ben Khelifa B, Salem B, El Kamel R. Neoplasm of a supernumerary undescended testis: A case report and review of the literature. Int J Surg Case Rep. 2018;53:345-7.
11. Noroozian J, Farishta D, Ballow D, Sonstein J, Orihuela E, Eyzaguirre E, Presentation C. Primary Malignancy in a Supernumerary Testicle Presenting as a Large Pelvic Mass. Hindawi Case Rep Urol. 2017;4529853.
12. Cosmo G, Silvestri T, Bucci S, Bertolotto M, Trombetta C. A singular case of polyorchidism. Archivio Italiano di Urologia e Andrologia. 2016;88:4.

Cite this article as: Valente T, Ferreira N, Ferreira G, Figueiredo A, Bárbara P, Aniceto C, et al. A rare case of polyorchidism in a 48-year-old man with inguinal hernia. Int Surg J 2022;9:2059-61.