CASE REPORT

Doppler ultrasound findings in a patient with primary infertility and triorchidism

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SUMMARY

Polyorchidism, presence of more than two testes in a scrotum, is a rare clinical entity. To date, about 200 cases have been reported in the literature. We report a case of triorchidism in a patient presenting with a 7-year history of primary infertility. Clinical examination revealed two completely separate testes with two separate epididymides and single vas suggesting a complete division of the genital ridge and partial duplication of the vas deferens. In our patient, the clinical picture together with ultrasound findings were characteristic enough to exclude other diagnoses, allowing conservative management. We also present the Doppler ultrasound findings of this rare condition, which may be helpful for comparison in patients with triorchidism who present with testicular torsion.

BACKGROUND

Presence of more than one testis in a hemiscrotum is known as polyorchidism. In the 14th century, the Pope allowed a man to marry twice because the man had three testes. Cases of three testes are usually attributed to spermatocoele,¹ but ours could have been a case of true triorchidism. Knowledge of this rare clinical entity is important as it may present as a scrotal mass and could be confused with extratesticular tumours by an inexperienced physician, leading to unnecessary investigations. We also report testicular blood flow detected on Doppler ultrasound in our case of incidentally detected triorchidism. These findings suggest that the blood flow of the extra testis is similar to that of the ipsilateral and contralateral testes.

CASE PRESENTATION

We report a case of a 30-year-old man who presented to us for evaluation of primary infertility. He was unable to conceive during his 7 years of marriage. He had regular unprotected sex without any intercourse-related problems. Medical and surgical history was unremarkable for surgery, orchitis, trauma and tuberculosis. Three of his brothers were married and fertile while one younger brother and one sister were unmarried. On examination, the patient had two ovoid, smooth testes palpable in the left hemiscrotum. Both testes in the left hemiscrotum had separately palpable epididymides on posterior aspect and a single vas deferens (figure 1). The right testis was normal and there was no clinical varicocele bilaterally.

INVESTIGATIONS

Semen analysis showed a sperm count of 5 million/mL with 20% progressive motility. Serum follicle stimulating hormone (FSH) was normal (7.22 IU/mL). Serum β -human chorionic gonadotropin (β hCG; <2 mIU/mL), alphafetoprotein (α FP) (0.6 IU/mL) and lactate dehydrogenase (LDH; 348 IU/L) were also found to be within normal limits. Scrotal ultrasound confirmed the clinical findings. There were two ovoid testes in the left hemiscrotum, completely separate from each other and measuring 22×16 mm each. Epididymides of both these testes were also separately visualised, and measured 7 and 6.6 mm, respectively (figure 2). The peak systolic velocities (PSVs) of the intraparenchymal and subcapsular branches of the right testicular artery were 3.81 and 4.23 cm/s, respectively (figure 3). The corresponding PSVs of the left upper testis were 5.78 and 4.65 cm/s; and for the left lower testis, were 3.95 and 4.09 cm/s, respectively (figure 4). These similar PSVs and lack of microlithiasis were suggestive of normal testicular parenchyma in all three testes.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis includes a scrotal mass. Benign conditions such as epididymal cyst, spermatocele and encysted hydrocoele are much more common than triorchidism. These are easily distinguishable from triorchidism by the presence of fluid, clinically by transillumination and confirmed using ultrasonography. Solid tumours are difficult to differentiate based on clinical and sonographic

Figure 1 Scrotal examination, two testes are visible in the left hemiscrotum.

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Figure 2 Ultrasound of the scrotum demonstrating two testes with two separate epididymides.

features. Particularly, extratesticular scrotal tumours may be difficult to distinguish from triorchidism. MRI is useful if the anatomy is not clear on clinical examination or if there is suspicion of an extratesticular scrotal tumour.² Splenogonadal fusion has been reported to mimic supernumerary testis³ but in our case the extra testis was easily distinguishable due to the presence of its own epididymis.

OUTCOME AND FOLLOW-UP

The patient is being managed conservatively for his infertility and has been advised fertility evaluation for his female partner. His oligoasthenospermia cannot be attributed to this morphological abnormality in the scrotum and no reversible factor could be identified. The couple is likely to be a candidate of in vitro fertilisation/intracytoplasmic sperm injection (IVF/ICSI). For IVF/ICSI, spermatozoa would be readily available from the patient's semen in this case.

We plan to follow-up the patient with clinical examination, semen analysis and ultrasonography after 3 months, and thereafter due to theoretical risk of malignancy in the supernumerary testis.

DISCUSSION

Cases of polyorchidism have been reported in humans as well as in animals such as dogs,⁴ and in hummingbirds.⁵ A meta-analysis published in 2009 identified 178 published cases

of polyorchidism with triorchidism being the most common form.⁶ The embryological origin of polyorchidism has been postulated to involve duplication or division of the genital ridge.⁷ Various classification systems have been suggested for this condition.⁸ All these classifications take into account whether the supernumerary testis share the vas, epididymis, or both. Various classification systems have been suggested for this condition.² Ninety per cent of cases reported in the literature are either type II (both testis share the epididymis and vas) or type III (both testis share a common vas). Our patient had a single vas and two palpable epididymides, which was also confirmed with ultrasonography. This was type III polyorchidism according to Leung⁹ and type A2 according to Bergholz *et al.*⁸

The clinical presentation of polyorchidism ranges from an asymptomatic mass to testicular torsion. Most cases in the literature are reported after surgical intervention for either an undescended testis or testicular torsion. Few cases of triorchidism are identified during infertility evaluation.^{10 11}

Ultrasound Doppler is a useful modality for the diagnosis of polyorchidism in asymptomatic patients as well in patients with an undescended testis or testicular torsion. Radiological features in an asymptomatic man have been reported by Yalçinkaya *et al.*¹²

The classic appearance is of a well-circumscribed, ovoid, scrotal mass with echo pattern identical to ipsilateral testis. In patients with testicular torsion of supernumerary testis the

Figure 3 Doppler ultrasound demonstrating intraparenchymal and subcapsular peak systolic velocities of the right testis.

Figure 4 Doppler ultrasound demonstrating intraparenchymal and subcapsular peak systolic velocities of the left upper and lower testes. Note that these velocities are similar to those of the normal right testis.

diagnosis may be tricky. Torsion of appendix testis is difficult to distinguish clinically. Doppler ultrasound is a useful modality for diagnosis in such cases. The diagnosis of torsion is based on lack of flow in an extratesticular structure, which is isoechoic to the ipsilateral testis.²

Other more common conditions should be excluded before making a diagnosis on radiological features only. Graif *et al*⁹ reported a case of a ruptured extratesticular epidermal inclusion cyst mistaken for torsion with triorchidism.

The diagnosis of polyorchidism is ideally confirmed with histopathology. However, as suggested by Bhogal *et al*¹⁴ and others,^{8 15} histopathology is only necessary if the diagnosis is in doubt or other diagnoses are being considered. Findings that mandate histopathology include presence of microlithiasis or ultrasound features suggestive of an extragonadal tumour. Radiological diagnosis is based on ultrasound or MRI. The only meta-analysis of polyorchidism found that 73% of reported cases were histologically confirmed and 27% were diagnosed by ultrasound or MRI. MRI is especially useful in cases where

ultrasound and clinical features are atypical (eg, intra-abdominal testis) or suggestive of malignancy.⁶

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REFERENCES

- 1 Hirsh A, Dean N, Mohan P, *et al*. Andrology: natural spermatocetes in irreversible obstructive azoospermia—reservoirs of viable spermatozoa for assisted conception. *Hum Reprod* 1996;11:1919–22.
- 2 Kumar K, Das D, Shivaraj. Triorchidism with torsion. *Ann Med Health Sci Res* 2012;2:199–201.
- 3 Carragher AM. One hundred years of splenogonadal fusion. *Urology* 1990;35:471–5.
- 4 Tamminen TM, Leinonen MR, Käck H, *et al*. A polyorchid dog. *Reprod Domest Anim* 2012;47:e26–8.
- 5 Witt CC, Bautista E. Triorchidism in a Hummingbird. *Wilson J Ornithology* 2011;123:632–5.
- 6 Bergholz R, Wenke K. Polyorchidism: a meta-analysis. *J Urol* 2009;182:2422–7.
- 7 Kheirandish P, Chingewundoh F. An unusual case of triorchidism. *JRSM Short Rep* 2010;1:55.
- 8 Bergholz R, Koch B, Spieker T, *et al*. Polyorchidism: a case report and classification. *J Pediatr Surg* 2007;42:1933–5.
- 9 Leung A. Polyorchidism. *American family physician* 1988;38(3):153–56.
- 10 Hassan A, El-Mogy S, Mostafa T. Triorchidism: a case report and review of similar conditions. *Andrologia* 2008;40:265–9.
- 11 Ahmadnia H, Molaei M. Polyorchidism in a patient with azoospermia. *Saudi J Kidney Dis Transpl* 2009;20:670–1.
- 12 Yalçinkaya S, Şahin C, Şahin AF. Polyorchidism: sonographic and magnetic resonance imaging findings. *Can Urol Assoc J* 2011;5:E84–6.
- 13 Graif A, Gakhal M, Iacocca MV, *et al*. Ruptured extratesticular epidermal inclusion cyst mimicking polyorchidism with torsion on sonography. *Emerg Radiol* 2014;21:643–5.
- 14 Bhogal RH, Palit A, Prasad KK. Conservative management of polyorchidism in a young man: a case report and review of literature. *Pediatr Surg Int* 2007;23:689–91.
- 15 Singer B, Donaldson J, Jackson D. Polyorchidism: functional classification and management strategy. *Urology* 1992;39:384–8.

Learning points

- Polyorchidism is a rare condition that may present with an asymptomatic testicular mass or be incidentally discovered in patients being evaluated for infertility or cryptorchidism.
- Clinical examination supplemented with ultrasound usually confirms the diagnosis.
- MRI or histological diagnosis is required if there is concern of malignancy.
- In infertile patients with polyorchidism, all functional gonadal tissue connected with the tubular system should be conserved.

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