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## LETTER TO THE EDITOR

Male Health

# Polyorchidism: the case in a young male and review of the literature

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Dear Editor,

Polyorchidism refers to the presence of more than two testicles. Blasius described it for the first time in 1670 as an incidental finding in an autopsy, while Lane was the one who performed the first histological description of a supernumerary testis in 1895.<sup>1,2</sup> Before imaging diagnosis techniques, this condition could only be verified under surgical exploration. Nowadays, current imaging techniques have increased the number of diagnosis of this anatomical variation. On the other hand, there is still a certain group of physicians who use histological analysis as the unique way to diagnosis.<sup>3</sup>

We report a 14-year-old male patient, with unremarkable medical or surgical history, who arrived to the emergency room with right testicular pain. The patient reported an acute testicular pain that awakened him at night and disappeared spontaneously. He did not report the presence of lower urinary tract symptoms or scrotal trauma. Physical examination revealed left and right testicles normal in size, position and firm in consistency. No varicocele was palpable on either side. Vasa, epididymis, and penis were unremarkable. In the right scrotum a 1 cm painless mass was palpable.

A scrotal ultrasound revealed a rounded structure of 1 cm of diameter with clear limits, located in the right hemiscrotum adjacent to the lower pole of the right testis and presenting the same echogenicity and Doppler intensity as the testis. Scattered micro-calcifications were seen in both testicles. Testicular tumor markers profile showed lactate dehydrogenase (LDH), alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin ( $\beta$ -hCG) in normal levels. Magnetic resonance imaging (MRI) confirmed the diagnosis with a left normal testicle with maximum diameters being approximately 3.5 cm and normal epididymis. Two testicles were identified on the right side; the larger one was approximately 3.2 cm, and a second one, completely independent testicle, measuring 1.35 cm long, was found adjacent to the larger one. Each testicle seemed to show its own epididymis. The presence of a 6 mm cyst on the head of the epididymis of the larger right testicle was also reported. In conclusion, diagnosis was polyorchidism with duplication of the right testicle (**Figure 1**).

The aetiology of polyorchidism is still unknown. Several theories have been proposed to explain the pathogenesis of such anomaly.<sup>2,4</sup>

Leung<sup>5</sup> created a four-group anatomical classification of polyorchidism based on testis embryology. Group one consists of polyorchidism with no vas deferens or epididymis in the supernumerary testis. Group two includes the testes that share these two mentioned structures with the ipsilateral testicle. The third is formed by testes with their own epididymis and sharing the vas deferens. Finally, group four corresponds to supernumerary testes with their own annexes.<sup>5</sup> Anatomical and functional classification proposed by Singer *et al.*<sup>6</sup> divides polyorchidism into two groups: group I, in which there is reproductive functionality, given that the tubules linking to the epididymis and vas are permeable. And group II, with infertile patients. In turn, both groups have two subgroups based on whether there is a scrotal location of the testis (IA, IIA) or an ectopic location (IB, IIB).<sup>6</sup>

Triorchidism is the numeric alteration most frequently found, as well as left laterality (60%) and the scrotal location (70%). Our case, though its scrotal location, is right sided. However, several locations have been described at any point along the way of embryological testicular descent to its final scrotal location.<sup>1–3,7</sup> According to the literature, patient age at diagnosis ranges from 15 to 25 years, with a mean age of 17 years in the study including the largest series reported.<sup>3</sup> Diagnosing this malformation in childhood or in individuals over 50 is even more infrequent.

The clinical signs and symptoms consist of scrotal pain, which may be intermittent and associated or not to palpation of scrotal tumor, as in the case we report.<sup>1</sup> Many patients have never experienced clinical symptoms, and the diagnosis might be incidental.<sup>7</sup> Imaging studies are essential for reaching the diagnosis. SU would disclose the supernumerary testes. However, it might be necessary to perform techniques such as MRI, which could provide diagnostic confirmation.<sup>8</sup> Some papers report that MRI does not provide additional information and recommend it should be kept for cases where intra-abdominal polyorchidism or malignancy is suspected.<sup>3</sup> On the contrary, other studies recommend that a pelvic MRI should be mandatory after SU.<sup>2,9</sup> In our case, MRI was helpful for confirmation of the anatomy. Hormone determination of  $\beta$ -hCG, AFP and LDH could be requested in case of doubt of testicular cancer.<sup>1</sup>

Regarding to associated pathologies, inguinal hernia (24%–30%), cryptorchidism (15%–40%), testicular torsion (13%–15%), hydrocele (9%), pain (7%), hypospadias, persistence of Müller ducts (7%–9%), chromosome alterations (3%), testicular cancer (6%) and varicocele (1%–1.4%) are possible.<sup>1–3,7,9</sup> Among all of these,



**Figure 1:** Magnetic resonance imaging showing the three testicles in the scrotum.

testicular cancer is the most remarkable, which occurs more frequently in cryptorchidism.<sup>3</sup> Another situation found in these patients is infertility that can lead to the diagnosis in some cases.<sup>3,7</sup>

Increased risk of testicular cancer (especially in cryptorchidism) has been the reason to perform surgery in these patients during the last years.<sup>9</sup> However, since the improvement in imaging techniques, most reports propose a close follow-up and decline an early invasive management.<sup>8,10</sup> In a recent meta-analysis, a management protocol was proposed based on whether the supernumerary testis was located in the scrotum or not. Nonscrotal location should be managed by orchiectomy. For the scrotal location, except symptoms requiring surgery or signs of malignancy in imaging studies, a conservative management with imaging techniques regularly performed could be proposed.<sup>3</sup>

Our patient has not scrotal pain in the follow-up; he has been instructed in testicular auto exploration and was being followed-up every 2 years with SU.

Despite the oddity of this malformation, it is important to consider it in the differential diagnosis of testicle cancer, given how different the management of these two pathologies is. The need for imaging diagnosis is clear. However, more accurate techniques than SU to diagnose and follow-up are seldom needed, unless malignancy or complications are suspected.

#### AUTHOR CONTRIBUTIONS

FB had full access to all the data in the study and took responsibility for the integrity of the data and the accuracy of the data analysis. AA and RR carried out the study concept, design, the acquisition of data and drafting of the manuscript. GM, EB, SL, JAB carried out the critical revision of the manuscript for important intellectual content and supervision.

#### COMPETING INTERESTS

All authors declare no competing interests.

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