Two halves of a whole: Bilobed testis case report and implications in management of a rare condition

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ABSTRACT
Bilobed testicle is an exceedingly rare congenital malformation with only seven cases reported in the literature. We describe the case of a 39-year-old man who presented with a left-bilobed testicle, resembling testicular malignancy. Despite its rarity, bilobed testicle should be considered in the differential diagnosis when a testicular mass is detected. Once malignancy has sufficiently been ruled out, bilobed testicle is typically managed conservatively.

Introduction
With only seven cases reported in the literature, bilobed testicle is a poorly understood and perplexing condition. Although the exact etiology of bilobed testicle has not been elucidated, it is postulated to be a subtype of incomplete polyorchidism - another rare congenital malformation with less than 200 cases reported. Embryologically, the etiology of polyorchidism is understood as aberrant division or multiplication of the genital ridge, likely due to a peritoneal band. Thus, bilobed testis likely arises from incomplete division of the genital ridge secondary to a peritoneal band.

Debate persists between providers whether bilobed testicle confers an elevated risk for sequelae such as torsion or malignancy. Advocates who argue that bilobed testicle is largely benign recommend conservative treatment; however, due to the lack of long-term studies and rarity of the condition, it is difficult to assess long-term outcomes. More broadly, polyorchidism is typically asymptomatic but is associated with several different pathologies including inguinal hernia, testicular torsion, cryptorchidism, infertility, and malignancy.

We describe a case of a bilobed testis in a 39-year-old man diagnosed by ultrasound imaging, the oldest such patient with this condition.

Case report
A 39-year-old man was referred to Urology clinic for evaluation of a left testicular mass. His history is significant for mild intermittent left testicular discomfort that dates back a year prior to presenting. This discomfort resolved after a course of antibiotics. Upon physical examination, testes were found to be descended into the scrotum bilaterally, and a mass was observed at the superior aspect of the left testicle (Fig. 1). Ultrasound revealed a left testicle with bilobed configuration measuring 6 × 2.3 × 3.3 cm with normal vascularity. Equivalent echogenicity was found bilaterally. The left epididymis was unremarkable. The patient was evaluated and found to have a left-sided grade II varicocele as well as small bilateral hydroceles. Due to the equivalent echogenicity of the testicular mass, conservative treatment and monthly self-examination was recommended with interval repeat imaging.

Discussion
Historically, due to the lack of imaging technology, diagnosis of polyorchidism was made incidentally with exploratory surgery. Current consensus is that ultrasound and MRI are sufficient to diagnose polyorchidism, including bilobed testicle, and to rule out malignancy. With our patient, we personally felt confident with the use of ultrasound to diagnose the bilobed testicle.

The most common complication of polyorchidism is testicular torsion with certain studies estimating the rate as high as 15% compared to the general population of 0.025%. As such, surgical management has not been indicated in the treatment of uncomplicated, asymptomatic bilobed testicle. Despite this, the risk for testicular torsion is still high and patients should be counseled on the signs and symptoms of sudden testicular torsion.

Debate persists as to whether bilobed testicle confers an increased rate of testicular malignancy. The malignancy rate in polyorchidism is an estimated 5.7%–7% compared to that of the general population rate.

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Despite the conferred increased risk of malignancy in polyorchidism, there have been no reported cases of malignancy in bilobed testicle; therefore, prophylactic surgical management has not been recommended. Furthermore, measuring tumor markers such as alpha-fetoprotein and beta-human chorionic gonadotropin has not been indicated. Conservative management of bilobed testicle stands in contrast to polyorchidism, where the risk of malignancy is much greater. In our case, the patient’s uncomplicated bilobed testicle with equivalent echogenicity would most likely benefit from a conservative approach with serial imaging and follow-up.

In summary, bilobed testicles should be suspected in the differential diagnosis when a testicular mass is detected. Malignancy and testicular torsion must first be ruled out, preferably with the use of ultrasound imaging. In uncomplicated cases, such as in our patient, a conservative approach is recommended with serial imaging and self-examination. Evaluation of long-term outcomes is necessary to further characterize the complication rates of bilobed testicle, though this is challenging given its rarity.

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Fig. 1. Scrotal ultrasonography revealed left testicle with clear division of superior and inferior poles of the testis, consistent with abnormal bilobed configuration.