Case Report

Unusual Combination of Bilateral Testicular Microlithiasis and Tubular Ectasia of the Rete Testis with Left Intra- and Extratesticular Varicocele in a 17-year-old Boy

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ABSTRACT: We describe the rare combination of testicular microlithiasis, unilateral intra/extratesticular varicocele, and tubular ectasia of the rete testis in a 17-year-old boy who presented with testicular pain following a trauma. He had a prior history of undescended testis and orchiopexy in childhood. His workup included a normal abdominal ultrasound and a sperm analysis demonstrating a low sperm count with sperm dysmotility. A follow-up ultrasound was unchanged, and he has been managed conservatively. This combined set of findings has not been previously reported. © 2011 Wiley Periodicals, Inc. *J Clin Ultrasound* **40**:314–318, 2012; Published online in Wiley Online Library (wileyonlinelibrary.com). DOI: 10.1002/jcu.20878

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Trauma and resultant pain are common indications for testicular sonography (US) in the pediatric population, especially in children participating in contact sports. In the trauma setting, US examination should be performed without delay to rule out emergencies such as testicular torsion or testicular rupture. Occasionally, nonneoplastic lesions unrelated to the trauma are found within the testis and it is essential for the radiologist to accurately diagnose these conditions. Cystic lesions include simple cysts, epidermoid cysts, tubular ectasia of the rete testis, intratesticular varicoceles and spermatoceles, and abscesses.[1] We present the case of a 17-year-old boy whose US examination revealed bilateral testicular microlithiasis, unilateral intra- and extratesticular varicoceles, and tubular ectasia of the rete testis. To our knowledge, this combination of findings has not been reported in a pediatric or adult patient.

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CASE REPORT

A healthy 17-year-old boy presented to the Emergency Department complaining of a 4-day history of left testicular, scrotal, and generalized groin pain that initially occurred after an injury during a football game. He had noticed bruising and swelling of his left scrotal sac after the injury, which had gradually been improving. He was born with an undescended left testis and underwent orchiopexy in infancy. High-resolution US was performed utilizing a Philips iU22 ultrasound system and L12–5 50-mm broadband linear array transducer (Philips Ultrasound, Bothell, WA). It revealed bilateral testicular microlithiasis in symmetrically sized testes (Figure 1). Within the left testis there were larger serpiginous tubular structures throughout the testicular parenchyma, in addition to an extratesticular varicocele (Figure 2). Color Doppler imaging demonstrated flow within the left-sided intratesticular varicocele (Figure 3). Spectral waveform analysis showed a venous flow pattern (Figure 4). Smaller caliber tubular structures

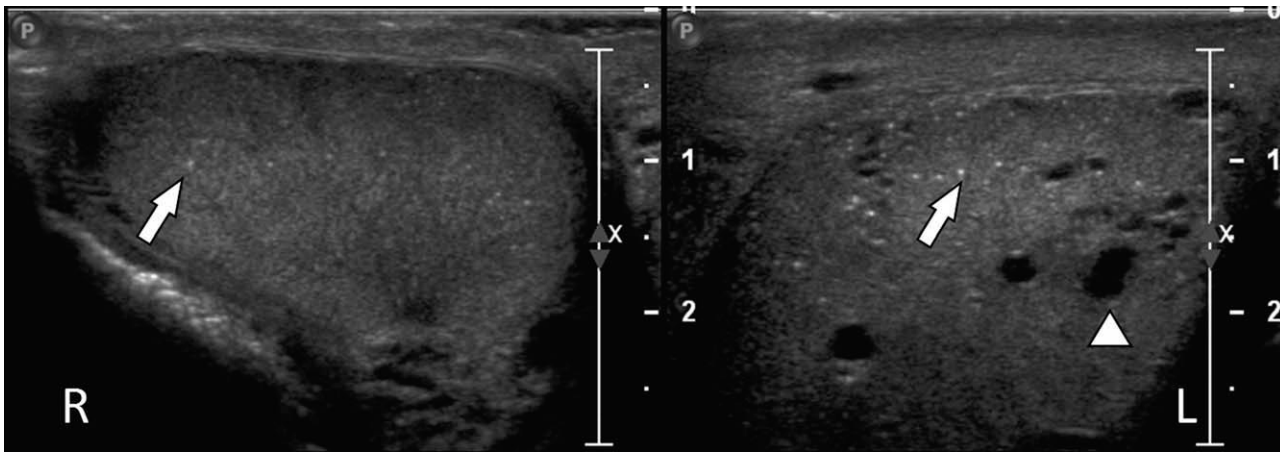

FIGURE 1. Longitudinal grayscale sonograms of the bilateral testes show nonshadowing punctate echoes consistent with microlithiasis (arrows) and anechoic structures within the left testicular parenchyma (arrowhead) representative of an intratesticular varicocele. L, left; R, right.

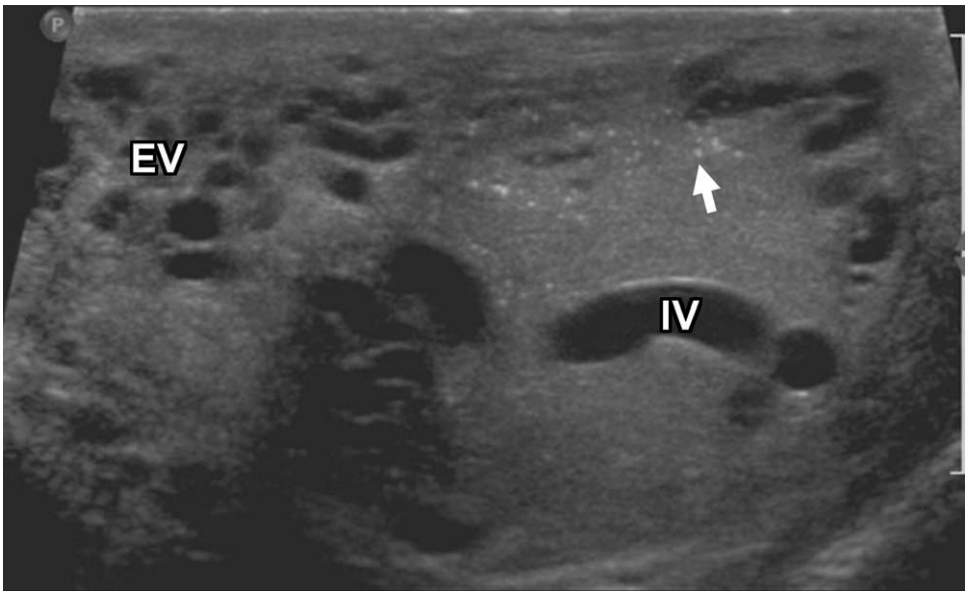

FIGURE 2. Transverse grayscale sonogram of the left testis shows microlithiasis (arrow). An extratesticular varicocele is present (EV), in addition to large anechoic structures within the testicular parenchyma representing an intratesticular varicocele (IV).

near the left testicular mediastinum demonstrated no internal flow, consistent with tubular ectasia of the rete testis (Figure 5).

After 6 weeks, the patient returned to the urology clinic and was completely asymptomatic. A repeat testicular US examination was unchanged. Given the rarity of this constellation of findings, there is no well-defined course of management. After a multidisciplinary conference, it was decided to first perform a renal US examination to evaluate for developmental anomalies, which can be associated with the testicular abnormalities described above. This examination was normal. Additionally, to further guide the decision of conservative versus surgical therapy, a sperm analysis was ordered to assess the impact of the varicocele, which revealed a low sperm concentration of 10 million/ml (normal 20–999 million/ml). Additionally, sperm motility, forward progression, and speed were all decreased. The patient is currently scheduled for follow-up to discuss potential surgical therapy.

DISCUSSION

This case demonstrates a combination of three nonacute intratesticular findings not previously described on testicular US. Testicular microlithiasis is defined as five or more echogenic nonshadowing foci less than 3 mm within the testicular parenchyma.[2] The etiology of this condition is unknown, but it may represent degeneration of

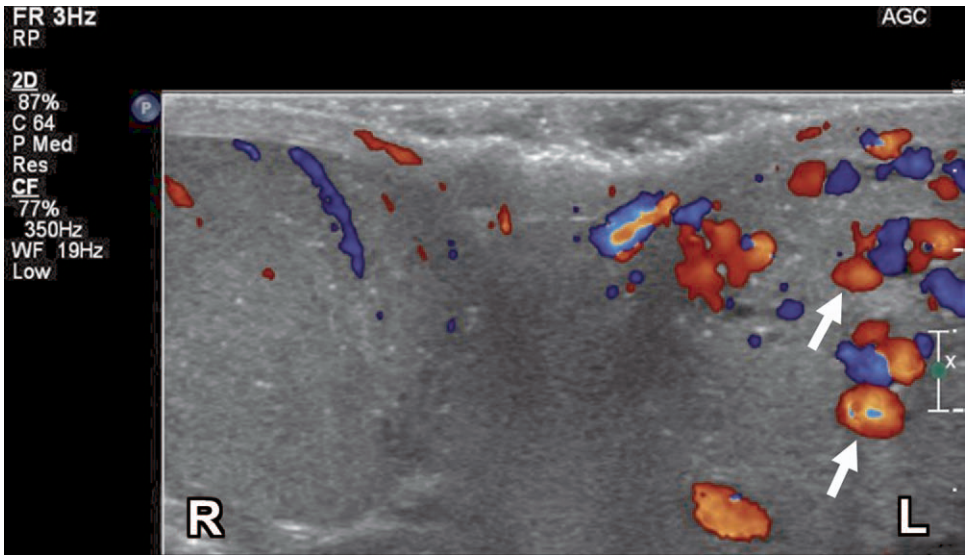

FIGURE 3. Transverse color Doppler sonogram of both testes demonstrates spontaneous flow within the larger left anechoic tubular structures (arrows), consistent with intratesticular varicocele.

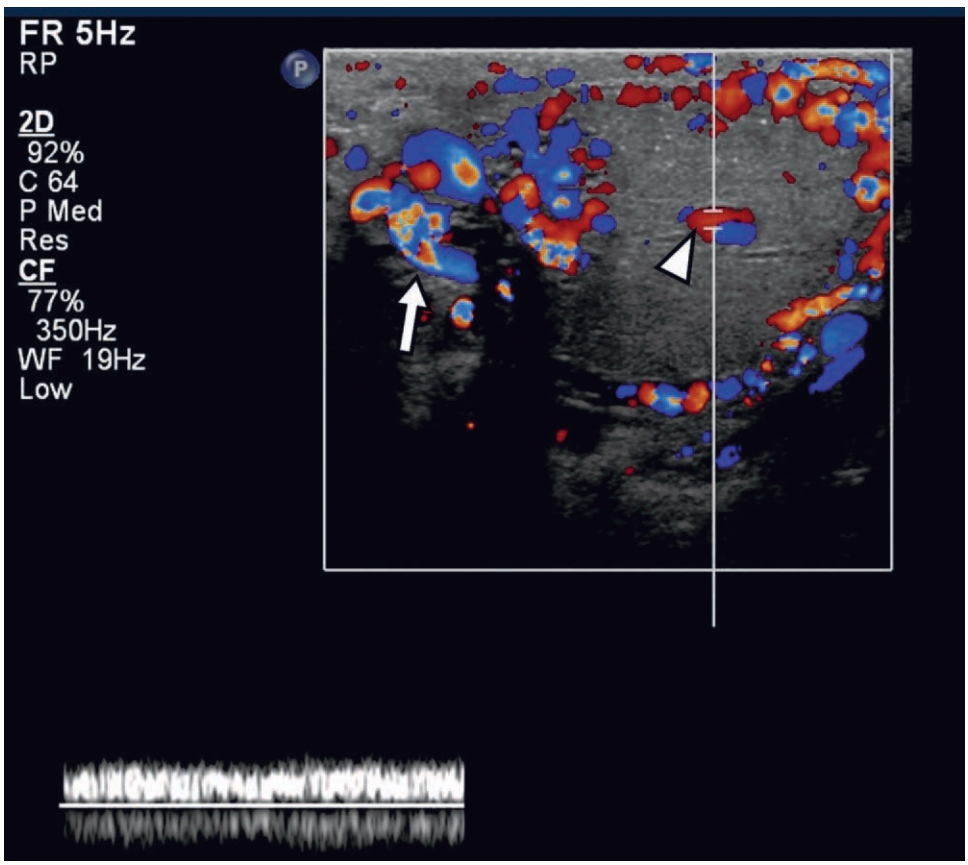

FIGURE 4. Spectral Doppler analysis of the left intratesticular varicocele (arrowhead) demonstrates a venous flow pattern. The extratesticular varicocele is also seen in this image (arrow). Both varicoceles slightly increased with Valsalva maneuver.

the seminiferous epithelium with subsequent debris accumulation within the tubular lumen.[3] It has been associated with a wide variety of benign and malignant pathologies including cryptorchidism, Klinefelter's syndrome, germ cell tumors, prior trauma, prior infection, and cystic dysplasia of the rete testis.[2–4] Testicular microlithiasis may be seen in up to 4.2% of asymptomatic boys aged 0–19.[2] There are no current consensus guidelines for the management and monitoring of these

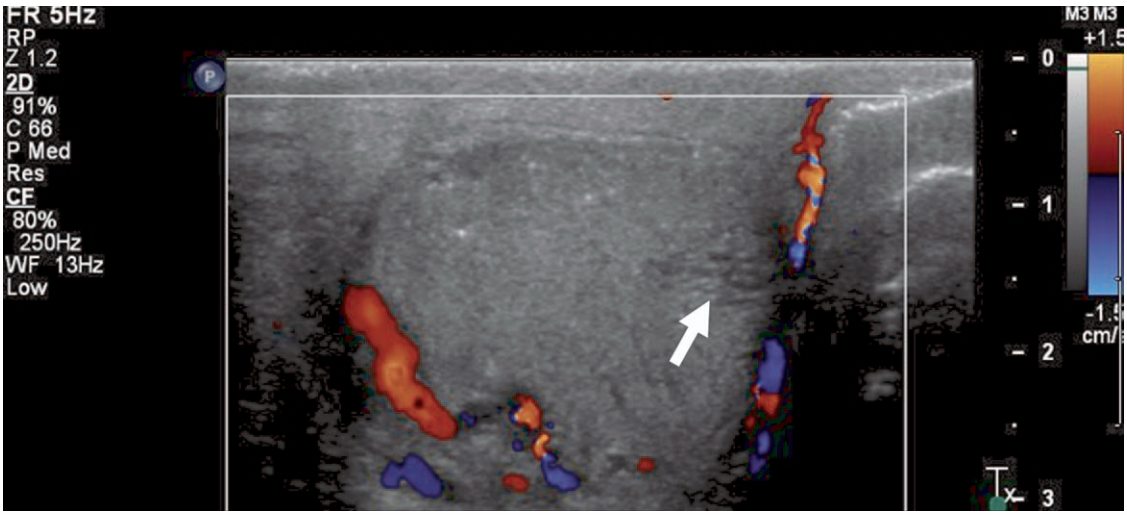

FIGURE 5. Transverse color Doppler sonogram of the left testis demonstrates anechoic tubular structures near the testicular mediastinum (arrow). These demonstrate no flow, consistent with tubular ectasia of the rete testis.

patients. Although some have advocated annual or biannual monitoring, there is no definitive evidence that this condition is preneoplastic in and of itself.

The second uncommon finding demonstrated in our case was tubular ectasia of the rete testis. This is most often encountered as an isolated finding; however, it has been reported in association with ipsilateral renal agenesis, multicystic kidney disease, renal dysplasia, undescended testis, seminal vesicle cysts, Potter's syndrome, vesicoureteral reflux, and absence of the vas deferens.[4,5,6] The rete testis is a network of tubules carrying sperm from the seminiferous tubules to the efferent ductules, which then drain into the epididymis. Tubular ectasia is hypothesized to occur when the efferent ductules become partially or completely blocked causing spermatozoa-containing cysts to form.[5] For others, it results from failure of the mesonephric structures to fuse with the testis. The condition is benign, with no malignant potential. Although it has been described in children previously, tubular ectasia is more commonly seen in men older than 50.[6] A predisposing condition (prior or chronic epididymo-orchitis, vasectomy, prior hernia surgery, etc) may be discovered in some cases.[6] The overall prevalence of tubular ectasia is thought to be around 5%. The true prevalence of this finding in the pediatric population is unknown.

Although extratesticular varicoceles are relatively common, our patient also had a less common intratesticular varicocele. The first cases were reported by Weiss et al in 1992 and were described sonographically as straight or tubular anechoic structures radiating from the mediastinum testis into the testicular parenchyma measuring at least 2 mm in diameter and demonstrating increased flow with Valsalva maneuver.[7] Overall, this lesion is rare with an unknown prevalence in the pediatric population. Less than 50 cases have been reported in the literature.[8] Up to one-third of cases may demonstrate bilateral involvement. The clinical consequences of extratesticular varicocele, namely infertility, are well established. There have been an insufficient number of cases reported to deduce the true significance of an intratesticular varicocele; however, at least one case of infertility has been reported related to an isolated intratesticular varicocele.[8] This may be due to differences of local temperature within the testis related to changes in hemodynamics. In the reported case, sperm count significantly improved after varicelectomy. Treatment of intratesticular varicocele is not well established, but the same endovascular embolization procedures as those used for extratesticular varicoceles have been tried with success.[8]

In summary, our reported case describes the coexistence of three relatively rare nonacute intratesticular findings. The patient had a prior undescended testis and orchipexy, which may have been a contributing factor. It is essential for the radiologist to accurately describe and diagnose these benign entities, despite their rarity.

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