Intratesticular varicocele: A rare finding of unknown significance. Report of 2 cases

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Abstract
Extratesticular varicocele is a frequent entity, with a reported occurrence of 20% in the male population. Conversely, intratesticular varicocele is an extremely uncommon condition, limited reported in the literature. The diagnosis cannot be made clinically and is established with Doppler ultrasound examination. Intratesticular varicocele may be associated with infertility, especially in cases of concurrent extratesticular varicocele. In this paper, we present two cases of young men who were diagnosed with both extratesticular and intratesticular varicocele.

Introduction
Extratesticular varicocele (ETV) is a common clinical condition, defined as the dilatation of the testicular vein and the pampiniform venous plexus within the spermatic cord, reporting in up to 20% of male population¹. The condition is considered congenital and is caused importantly by incompetent valves of the internal spermatic veins². Rarely, the condition occurs within the testis, so called intratesticular varicocele (ITV) and appears as dilated veins radiating from the mediastinum into the parenchyma³. Gray scale scrotal ultrasonography demonstrates tubular or serpentine vascular structures, which exhibit internal blood flow and positive Valsava maneuver⁴. In this paper, we present two cases of intratesticular varicoceles in young adults. In both cases, the condition was successfully resolved via varicocelectomy of the contemporary extratesticular varicocele.

Case presentation
A 23-year old man presented to our department re-

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questioning routine scrotal examination, as he had undergone left orchidopexy due to undescended left testicle when he was infant. Clinical examination of the scrotum showed a hypotrophic left testis, while the right testis was palpated normal. Scrotal ultrasonography confirmed the discrepancy of volume between testicles and dilated spermatic veins of left pampiniform plexus associated with retrograde blood during Valsava maneuver. In addition, a hypoechoic tubular and a cystic structure were found in mediastinum and lower pole of left testis, accordingly. During Doppler ultrasonography, these structures demonstrated constant internal blood flow which was exacerbated during Valsava maneuver (Figure 1, 2). Thus, a diagnosis of extratesticular and intratesticular varicocele in left testis was established. The right testis did not carry any evidence of varicocele or other pathology. A spermodiagram was requested which demonstrated oligo-asthenospermia and abnormal morphology. Due to severe oligospermia (approximately 5 millions/ml of ejaculation fluid), the patient was undergone endocrinologic setup, karyotype analysis and test for detection of deletions chromosome Y; all tests were normal. Under these circumstances, the combination of extratesticular and intratesticular varicocele was considered as the remaining treatable cause of sperm parameters deterioration; the patient was counseled to undergo subinguinal left varicocelectomy.

A second, 26-year-old patient, presented to our de-
partment reporting a subacute, dull, scrotal pain during the past 6 months. The patient reported scrotal trauma in his left testicle when he was a child; the condition was managed conservatively. During clinical examination bilateral large varicoceles were found; the lesions were visible and palpable at rest. Scrotal ultrasonography manifested bilateral, dilated extratesticular veins, outnumbered in left testicle, of maximal diameter of 5.4 mm, associated with retrograde blood flow during moderately tense inspiration. In addition, an intratesticular varicocele was found in the lower pole of left testicle (Figure 3, 4). The left testicle was found hypotrophic, but the right was measured within normal limits. A spermodiagram was requested which demonstrated deterioration of semen parameters. The patient was informed of the possible detrimental effect of clinical varicoceles on spermatogenesis and endocrine function of the organ and he was counseled to undergo bilateral varicocelectomy.

Postoperative follow-up was done in both patients, showing disappearance of extra- and intratesticular components of varicoceles. In the second patient, the scrotal pain was successfully regressed. After 10 months, an improvement of semen parameters regarding total sperm count, mobility and morphology was observed in both patients.

Discussion

In search of intratesticular varicocele in the medical literature, we came before an extremely rare entity. Since first description by Weiss et al, who noted intratesticular varicocele in two men with scrotal pain, only sporadic single case reports and small series have contributed to our knowledge of the condition. However, if a prevalence has to be estimated, the lesion does not seem to exceed 2% of cases of scrotal pathology.

The pathogenesis of intratesticular varicocele seems to be similar to the extratesticular component, as far as the lesion is found commonly in association with an extratesticular varicocele and shares some characteristics, like left predominance and increased flow during Valsava maneuver. In addition, previous ipsilateral genitourinary surgical procedures and especially orchiopexy may be implicated in etiology of both left and right ITV's, as has been reported in some case series. In the largest relative study, Meij-de Vries et al analyzing 105 patients who had prepubertaly undergone orchiopexy found a remarkably high prevalence (8.6%) of intratesticular varicocele in these men; a rational explanation given by the authors is the damage of testicular vessels during mobilization of the organ which may result in total incompetence of the valves of veins of the pampiniform plexus. For similar reason, isolated ITV may metachronously occur, as a result of testicular surgery for tumors. Testicular atrophy of various reasons has also hypothesized as a cause for the development of ITV, as the loss of surrounding parenchyma may allow enlargement of intratesticular venous structures. The latter hypothesis could also apply in our patients. Namely, it is possible that testicular atrophy might have come before, as a late complication of previous surgical exploration and trauma accordingly. As a lesion in anatomical continuity with the extratesticular component, the intratesticular varicoceles may have gradually developed as a subsequent phenomenon. To our experience, we cannot directly hold extratesticular varicoceles accountable for the development of ITV, as far as the condition has never been observed before in a large population of patients with both clinical and subclinical varicoceles managed in our center.

Since non-palpable during physical examination, the diagnosis of intratesticular varicocele is based on specific ultrasonographic findings. These lesions appear as tubular, serpentine or oval intratesticular structures which demonstrate increased and retrograded internal blood flow during Valsava maneuver. The lesions may be located within the parenchyma, in the mediastinum, or have subcapsular location. A minimum limit of 2 mm or greater in diameter may be used for the definition of an ITV; however, some authors define as ITV any intratesticular venous structure which exhibits reflux during Valsava, regardless of cutoff of 2 mm. These lesions are mainly on the left side; though bilateral or isolated right cases have been observed. The differential diagnosis of the condition consists of tubular or cystic lesions of the testis, like intratesticular, subcapsular cysts and tubular ectasia or cystic dysplasia of rete testis. Cystic teratoma of the testis is also a rare tumor which can mimic ITV, but a positive response to Valsava maneuver differentiates the condition.

The clinical spectrum of ITV may vary from asymptomatic course to painful scrotum, hypogonadism and infertility. The latter is possibly the most significant manifestation of ITV, arising likely from the concur-
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Conflicts of interest
The author declared no conflict of interest.

Abbreviations
EXT = extratesticular varicocele
ITV = intratesticular varicocele
CDU = colour doppler Ultrasonography
FSH = follicle-stimulating hormone

References