

Regional distribution of intratesticular varicoceles accompanying extratesticular varicocele cases

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ABSTRACT

OBJECTIVE: Only few cases of intratesticular varicocele (ITV) have been reported in the literature, mostly as single cases or a case series. In this study, we aimed to describe the topographic distribution of ITVs in the testicular parenchyma and the correlation between ITV accompanying extratesticular varicocele (ETV) and age.

METHODS: Approximately 10,537 patients with scrotal pain and swelling who visited our hospital between August 2014 and May 2020 and underwent scrotal colour Doppler ultrasonography were included in this study. Of these patients, 2285 had ETV, and the remaining patients (n=8252) had other testicular pathologies such as acute-chronic torsion, testicular mass, orchitis, significant testicular atrophy, or isolated extratesticular varicocele, and were thus excluded from the study. Among the 2285 patients, only 20 had ITV.

RESULTS: Of the 20 patients with ITV, 2 had isolated ITV, and the remaining had ITV accompanying ETV, of whom only one had a right ETV. One patient had left testicular atrophy accompanying an isolated ITV. Among the ITV cases, subcapsular ITV was the most common (n=13), followed by central ITV (n=4) and subcapsular and hilar ITV (n=3). The patients' ages ranged from 18 to 30 years.

CONCLUSION: ITV is a rare clinical entity that occurs only in young men, with most cases accompanying ETV. ITV is generally peripherally located in the testicular parenchyma. In this study, no correlations were found between ITV, ETV, and age. As in this study, testicular atrophy is rarely observed in patients with ITV. No studies have presented ITV cases with spermogram analysis results. Thus, the effect of ITV on male reproductive fertility is still unknown, and further study on this topic is needed.

Keywords: Intratesticular varicocele; scrotal Doppler; testis.

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Varicocele is a common cause of scrotal pain and infertility in the male population. In the pathophysiology of varicocele, increased pressure in the pampiniform plexus (PP) is the main etiologic cause [1–3]. In the PP, reverse venous flow towards the testis occurs because of increased pressure and reflux. The pressure increase causes an increase in PP diameter in the later stages and venous congestion in the testis. Thus, testicular pain and scrotal swelling due to the congestion, infertility due to

low sperm quality and count, and venous enlargement in the spermatic cord consequently occur. Although patients are generally asymptomatic, they may present to urology clinics because of pain, swelling, or infertility [4–6].

Physical examination and scrotal colour Doppler ultrasonography (S-DUSG) are essential in the diagnosis of varicoceles. Intratesticular varicocele (ITV) is extremely rare, as most cases of varicoceles almost always present as extratesticular varicoceles (ETVs). However,



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most ITV cases accompany ETV. The basic imaging method for the diagnosis of varicocele is S-DUSG. An increased PP diameter can be observed on S-DUSG. In varicoceles, the main S-DUSG finding is dilated tubular-shaped PP around the testis, with a reversed flow pattern (reflux). Most cases of varicocele are detected as ETV. On S-DUSG, ITV presents as dilated, tortuous, tubular venous vascular structures in the testicular parenchyma [7–9].

Only few cases of ITV have been reported in the literature, mostly as single cases or a case series [7–12]. Owing to the rarity of ITV, no study has investigated its relationship with infertility. Moreover, no research has examined the topographic distribution of ITV cases. Therefore, in the present study, we aimed to describe the topographical distribution of ITV cases and any correlation between ITV accompanying ETV and age.

MATERIALS AND METHODS

Patient Group Construction

Approximately 10,537 patients with scrotal pain and swelling who visited Eskisehir Yunus Emre State Hospital, Department of Urology outpatient clinic between August 2014 and May 2020 and underwent S-DUSG were included in this study. Of these patients, 2285 had ETV, and the remaining patients ($n=8252$) had other testicular pathologies such as acute-chronic torsion, testicular mass, orchitis, significant testicular atrophy, or isolated extratesticular varicocele, and were thus excluded from the study. Among the 2285 patients, only 20 had ITV. In this study, we included only ITV cases to define the topographical distribution of ITV cases. Owing to the unavailability of spermogram analysis tests in our hospital, patients were referred to a third-step hospital for spermogram testing. Hence, we could not evaluate the spermogram results of the patient group. This study was conducted in accordance with the Declaration of Helsinki. The Eskisehir Osmangazi University Non-interventional Clinical Research Ethics Committee granted approval for this study (date: 16.05.2023, number: 2022-245/02).

Ultrasonographic Evaluations

All ultrasonographic examinations were performed using the same machine (GE Logic P5, the Netherlands) with a 10- to 12-Mhz linear transducer by a general radiologist (Y.A.) who had 17 years of experience in the field of scrotal grayscale and Doppler USG. ETV was diagnosed

Highlight key points

- Most ITV cases were peripherally located in the testicular parenchyma.
- No correlations were found between ITV, ETV, and age.
- Testicular atrophy was rare among the ITV cases.

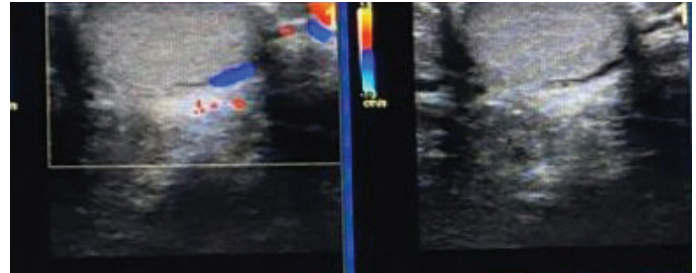


FIGURE 1. Peripheral ITV: An 18-year-old male patient was admitted to the urology outpatient clinic of our hospital with a complaint of left testicular pain. The scrotal Doppler USG performed in the USG outpatient clinic revealed tubular varicose venous enlargements in the peripheral subcapsular area of the left testis.

when the PP diameter was >2 mm on colour S-DUSG, and ITV was diagnosed when a dilated venous vascular structure was found in the testicular parenchyma. To distinguish the ITV from the testicular artery, the general radiologist used pulse-wave DUSG. The patients were assessed for PP reflux and diameter changes in an erect resting position using S-DUSG. In all the patients, the PP diameter was measured on S-DUSG. When an ITV was detected in the scrotal assessments, its diameter and reflux were recorded. To define the topographical distribution of ITV cases, central and peripheral localisation classifications were accepted as descriptive criteria. The testicular medulla and subtunical area were considered the central and peripheral locations, respectively. Testicular and epididymal volumes, echogenities, and peritesticular effusions were also evaluated.

Statistical Analysis

The distribution of each continuous variable was tested for normality using the Shapiro-Wilk test. The results are presented as mean \pm standard deviation (SD). Regression and Pearson correlation coefficient analyses were performed to determine the relationship between ITV and ETV. P values <0.05 were considered statistically significant. All analyses were performed using the SPSS version 25.0 software (SPSS Inc., Chicago, IL).

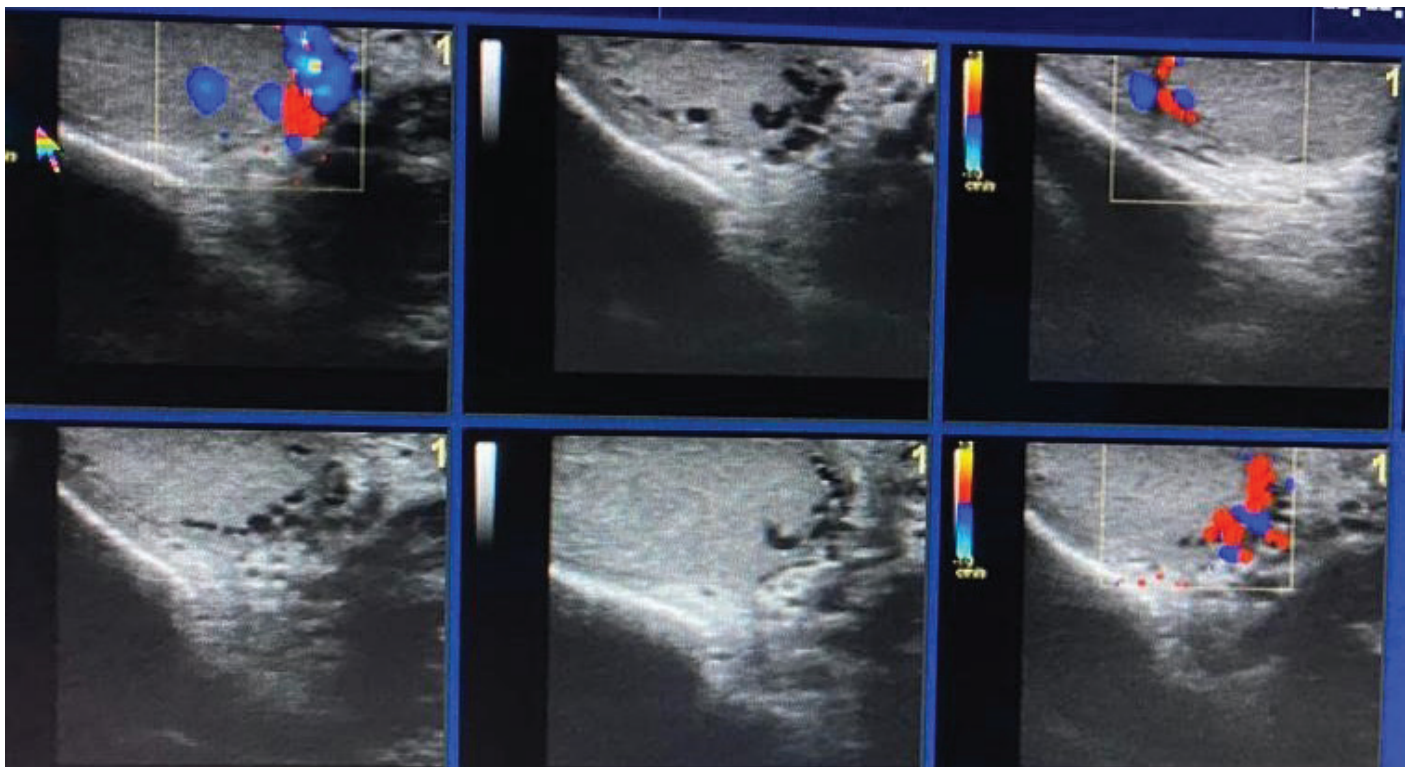


FIGURE 2. ETV+Peripheral subcapsular ITV: A peripheral subcapsular tubular ITV in a patient with a left extratesticular varicocele.

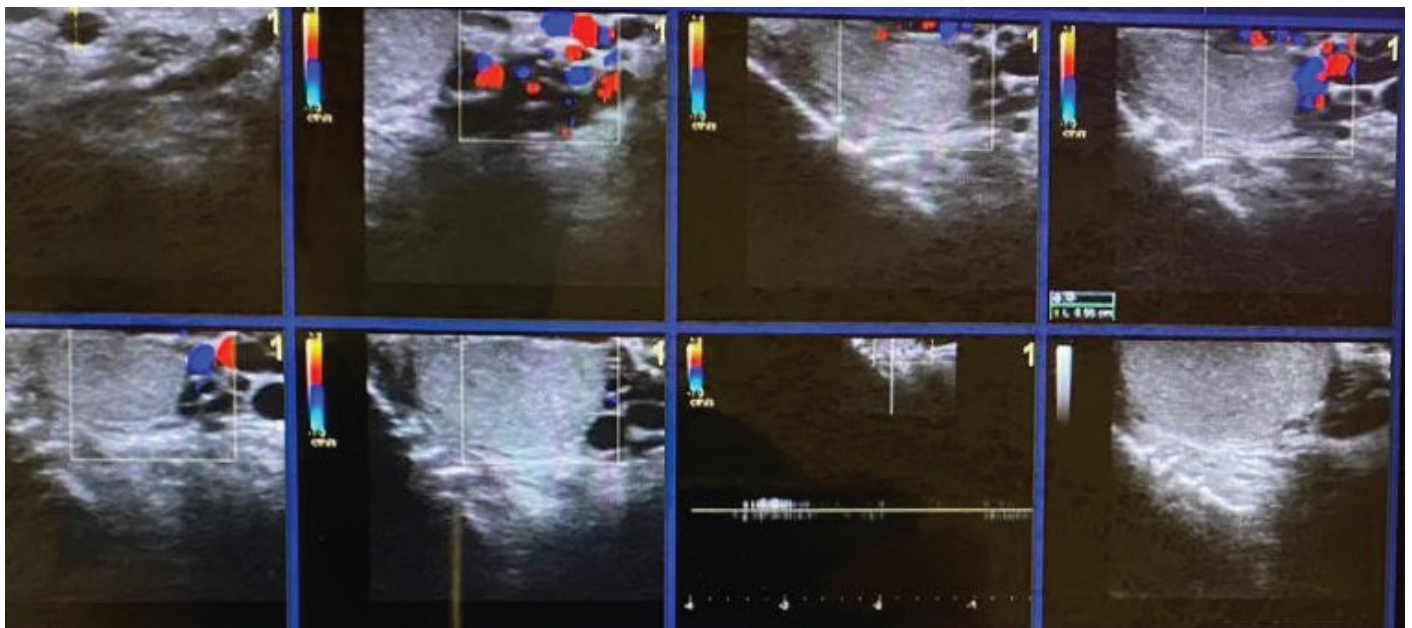


FIGURE 3. ETV+Peripheral subcapsular ITV: A case of subcapsular peripheral tubular ITV accompanying a left ETV in a 23-year-old patient who visited the USG outpatient clinic with a complaint of left testicular pain.

RESULTS

Among the 10,537 patients, 2285 had ETV, of whom 18 had an accompanying ITV. Two patients had an isolat-

ed ITV (Fig. 1), while the remaining patients had ITV accompanying ETV (Fig. 2, 3), with only 1 case of right ETV. Left testicular atrophy accompanying isolated ITV was observed in 1 patient. Among the patients with ITV,

TABLE 1. Demographics and imaging findings of the patients (n=20) with ITV

Age (years)	24±3 (18–30)
ITV diameter (mm)	1.72±0.85 (0.5–3.6)
ETV diameter (mm)	4.03±1.41 (1.8–6.2)
Ipsilateral ETV (%)	90
Contralateral ETV (%)	5
Central ITV (%)	20
Peripheral ITV (%)	80
Testicular hypoplasia (%)	18

ITV: Intratesticular varicocele; ETV: Extratesticular varicocele.

subcapsular ITV was the most prevalent (n=13), (Fig. 1–3) followed by central ITV (n=4) and subcapsular and central ITV (n=3). Table 1 presents the demographic and S-DUSG imaging findings. The patients' mean (\pm SD) age was 24±3 years (range, 18–30 years), ITV diameter was 1.72±0.85 mm (0.5–3.6 mm), and ETV diameter was 4.03±1.41 mm (1.8–6.2 mm). All the patients were young adults and had a history of walking or standing for long hours daily as soldiers performing compulsory military service in our city. Most patients had grade 2 ETV with ITV. Among all the patients, the prevalence of ITV was 0.2%, of which 0.9% were accompanied by ETV.

In this study, a group correlation analysis between ITV, ETV, and age was performed. No correlations were found between the three variables, as shown in Table 2. These results may be caused by our small sample size due to the rarity of ITV. For our patient group, we performed a graphical analysis of ITV and ETV (Fig. 4), which revealed a linear proportion between ETV and ITV.

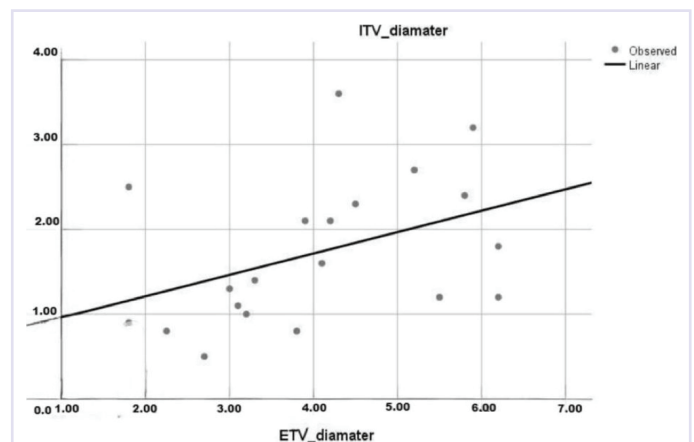
DISCUSSION

Approximately 10,537 patients with scrotal pain and swelling who visited our hospital between August 2014 and May 2020 and underwent S-DUSG were included in this study. ITV was detected in 20 patients of these patients, which accompanied a right ETV in 1 patient and a left ETV in the rest of the patients. Left testicular atrophy accompanying isolated ITV was also observed in 1 patient. Among the patients, subcapsular ITV was the most prevalent (n=13), followed by central ITV (n=4) and subcapsular and central ITV (n=3). The patients' ages ranged from 18 to 30 years.

TABLE 2. Correlation analysis between ITV, ETV, and age (n=20)

	Age	ITV_ diameter	ETV_ diameter
Age			
Pearson correlation	1	-0.213	-0.048
Sig. (two-tailed)		0.367	0.841
ITV_diameter			
Pearson correlation		1	0.416
Sig. (two-tailed)			0.068
ETV_diameter			
Pearson correlation			1

ITV: Intratesticular varicocele; ETV: Extratesticular varicocele.

**FIGURE 4.** Graphical analysis of correlation between ETV and ITV: Graphic shows direct proportion between ITV and ETV.

ETV is a common clinical condition that can affect approximately 15% of the male population, with a potential to cause scrotal pain and infertility. In severe cases or cases with abnormal spermiogram analysis results, surgical treatment might be required. In contrast to ETV, ITV is rarely encountered in daily clinical practice. It is characterised by enlarged venous varicose structures in the testicular parenchyma and almost always accompanies ETV, often involving the left testis. Right testicular involvement and isolated ITV are rare clinical conditions. Weiss et al. first reported varicose enlargements in the mediastinum testis. In addition, varicoceles have been reported as single cases or a case series in the literature [7–12].

Dhamija et al. [6] reported a case of isolated ITV in a 22-year-old patient with infertility. In their case report,

Kishore Kumar et al. [8] verified that ITV was associated with ETV. Pourbagher et al. [7.] verified that ITV is a rare entity that can cause scrotal pain and that varicose dilatation in the mediastinum testis might be observed on S-DUSG. The authors reported a PP diameter >2 mm and reflux flow in the PP in 2 cases.

Das et al. [13] conducted the first study on ITV, in which they detected ITV in 18 patients. They detected ITV on the right side in 12 of 25 testes and isolated ITV in 14 patients. Tubular ITV was detected in 20 patients; and oval-shaped ITV, in 5 patients. In contrast to their study, our study reports ITV cases in the left testes, almost all of which accompanying ETV. We think that this difference between the previous and present studies was caused by the inhomogeneity of our patient group, the racial differences between our patients, or our small sample size. Nevertheless, the tubular ITV morphology in all our cases is consistent with previous reports in the literature. In our study, the prevalence of ITV was 0.2%, of which 0.9% accompanied ETV.

Bucci et al. [14] detected 7 asymptomatic ITV cases in 342 S-DUSG images in their study, and almost all cases occurred in the left testis. A volume difference was found between the left and right testes. In contrast to this study, our study shows no volumetric difference between all patient groups except in 1 patient with testicular atrophy. Kessler et al. [15] found that in 12 male patients with ITV, ITV showed in 15 cases and occurred in the left testis in 7 cases, in the right testis in 2 cases, and bilaterally in 3 cases. The authors detected a subcapsular ITV in 60% of the cases and central or mediastinal ITV in 40%. Of the ITV cases, 26% were oval-shaped, and 26% were tubular. Oval-tubular shaped varicose enlargement was detected in 48% of the cases. In a study consisting of 5 patients, Atasoy and Fitoz [16] found a bilateral ITV in 2 patients and a left ITV in 3 patients. The ITV diameter was >2 mm in 3 patients and <2 mm in the rest of the patients. ITV accompanied ETV in almost all the patients, and varicose enlargement of the mediastinum testis was generally detected. Subcapsular localised varicose veins were rarely detected. In our study group, most ITV cases were subcapsularly localised, and all the patients had a tubular ITV. We verified that the ITV diameter was between 0.5 and 3.6 mm, and all cases showed reflux flow in the PP on pulsed-wave DUSG. In the correlation analyses were performed, we found no correlations between ETV, ITV, and age.

Our study has some limitations. First, no comparison could be made between spermiogram analysis results because of the unavailability of a spermiogram analysis in our hospital. Second, the sample size was small. Third, control S-DUSG was not performed after surgical treatment in any of the patients.

Conclusion

ITV is a rare clinical entity that occurs only in young adult men. Most ITV cases accompany ETV. Generally, ITVs are peripherally located in the testicular parenchyma. In this study, no correlations were found between ITV, ETV, and age. Testicular atrophy was found in few patients with ITV. No previous studies have reported ITV cases with spermiogram analysis results. Thus, the effect of ITVs on male reproductive fertility remains unknown, and further study on this topic is needed.

Ethics Committee Approval: The Eskisehir Osmangazi University Non-interventional Clinical Research Ethics Committee granted approval for this study (date: 16.05.2023, number: 2022-245/02).

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