Testing the undescended testis

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High prevalence of intratesticular varicocele in a post-orchidopexy cohort

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ABSTRACT

Background
Intratesticular varicocele (ITV) is an uncommon sonographic finding. A prevalence of up to 2% has been reported in men with testicular problems. In a cohort of men who had undergone prepubertal orchidopexy for acquired undescended testis, several cases of ITV were found. The aim of this study was to analyze the prevalence and clinical aspects of ITV in this cohort.

Methods
In a long-term follow-up study of position and growth of undescended testis after prepubertal orchidopexy, ultrasonography was used to identify men with ITV. Data on clinical presentation, testicular volume, and the location, size and Doppler aspects of intratesticular varicocele were collected and analyzed.

Results
Of the 105 men, 9 were identified with ITV (8.6%). In all patients, the side of orchidopexy correlated with the side of the ITV, and all were left-sided. The testis with ITV had a smaller volume than the testis without ITV (p = 0.026).

Conclusion
A remarkably high prevalence of ITV (8.6%) was found as well as a smaller volume of the testes with ITV in a cohort of men who had undergone prepubertal orchidopexy for acquired undescended testis.
EXTRATESTICULAR VARICOCELE IN ACQUIRED UNDESCENDED TESTES

INTRODUCTION

Extratesticular varicocele (ETV) is a common condition, occurring in approximately 10-15% of males. It is an abnormal dilatation of the veins of the spermatic cord, caused mostly by incompetent valves in the internal spermatic veins. Intratesticular varicocele (ITV) was first described in 1992 by Weiss et al, who described two patients with dilated intratesticular veins: straight or serpentine hypoechoic structures within the mediastinum testis and radiating into the testicular parenchyma. Since 1992, more cases of ITV have been reported. The reported prevalence is 0.4–2.0% in patients referred for scrotal ultrasound due to various testicular problems or for routine andrological evaluation. In a long-term follow-up study on the position and growth of acquired undescended testis after prepubertal orchidopexy, several cases of ITV were found. The aim of this study was to analyze the prevalence, clinical aspects and testicular volumes of the patients with ITV in this study cohort.

METHODS

From 1986 to 1999, 335 boys underwent prepubertal orchidopexy for acquired undescended testis at our hospital. In 2010, we conducted a follow-up study on testis position and growth. All 335 were requested by post to participate in this long-term evaluation. If no reaction followed, a second letter was sent and if still no reaction occurred, the patient was contacted by telephone. Patients were excluded if one or more of the following criteria was present in their medical history: recurrent epididymitis, chromosomal or hormonal abnormalities, hormonal medication, earlier orchidopexy or other inguinal surgery, congenital UDT, presence of a testicular germ cell tumor. 122 (36.4%) Patients gave written informed consent. 17 were excluded due to recurrent epididymitis (n=1), pubertas tarda and mental retardation (n=1), previous orchidopexy (n=2), inguinal hernia surgery (n=10), congenital UDT (n=2) and an immature teratoma (n=1).

Consequently, 105 (31.3%) patients were included. The age at orchidopexy of the patients was 2.4–13.9 years (mean ± SD; 9.2 ± 2.8 years). Of these 105 patients, 73 had undergone unilateral orchidopexy (33 left- and 40 right-sided), whereas 32 had undergone bilateral orchidopexy. As a result, 137 orchidopexied testes were studied.
follow-up, the patients’ ages ranged from 14.0 to 31.6 years (mean 25.7 years, SD ± 3.3). Examination at follow-up included scrotal sonography of both testes. If participants appeared to have findings suspicious for ITV, informed consent to participate in this study was asked. If consent was given, colour Doppler ultrasound was performed to confirm the diagnosis of ITV and to collect additional data.

**Definitions**
An undescended testis was defined as a testis which could not be manipulated into a stable scrotal position in its most caudal position, and further traction on cord structures was painful. It included high scrotal, inguinal or impalpable forms. An acquired undescended testis is an undescended testis which had been descended earlier in life and for which a previous scrotal position had been documented at least twice.

Orchidopexy was started with an inguinal incision. Subsequently, exploration of the groin took place and, if present, the open processus vaginalis was separated from the cord structures and ligated. Retroperitoneal funiculolysis and separation of the cremaster muscle was performed to mobilize the cord. Finally, the testis was fixated scrotally by making a scrotal incision and placing it in a created dartos pouch. All orchidopexies were performed under general anaesthesia as an outpatient procedure. An ITV is an anechoic tubular, oval or serpentine intratesticular structure that shows increased flow or reflux during a Valsalva manoeuvre.

**Questionnaire**
Participants were asked about their medical history. In addition, clinical symptoms such as testicular pain, scrotal swelling, epididymitis or gynaecomastia were scored. Moreover, the questionnaire included questions regarding fatherhood.

**Ultrasound**
The scrotal ultrasounds in the orchidopexy follow-up study were performed with a 12 MHz linear array transducer and Falco Auto Image (Falco Software Co, Tomsk, Russia). If findings were suspicious for ITV, a second sonographic examination was performed, with the UST-567 linear probe, frequency range 6-13 MHz, 50mm on the Aloka ProSound ALPHA 10 (Tokyo, Japan). Examinations comprised both B-mode and colour Doppler sonography. Before and during the Valsalva manoeuvre, the ITVs were scored
Intratesticular varicocele in acquired undescended testes

for diameter and location (mediastinum / parenchyma / subcapsular). Furthermore, the presence of ETV was assessed. Testicular volume was calculated by measuring the maximum length, width and height, using the formula for an ellipsoid = \( \pi/6 \times \text{length} \times \text{width} \times \text{height} \).

**Data analysis**

All data were collected and analyzed using SPSS, version 14.0. The independent t-test and the Mann-Whitney test were performed to calculate the differences in age and volume. A p-value of less than 0.05 was considered as statistically significant.

**RESULTS**

Of the 105 participants, 9 (8.6%) were identified with ITV. The characteristics of these 9 patients with ITV can be found in Table 1. Figures 1 and 2 show the colour Doppler ultrasound images of the ITV.

**Table 1** Data on 9 men with ITV found in a cohort of men after prepubertal orchidopexy for acquired undescended testis.

<table>
<thead>
<tr>
<th>patient number</th>
<th>age (years)</th>
<th>age at ORP (years)</th>
<th>side of ORP</th>
<th>side of ITV</th>
<th>clinical signs</th>
<th>dm ITV (mm)</th>
<th>location ITV</th>
<th>ipsi lateral ETV</th>
<th>volume testis ITV+ (ml)</th>
<th>volume contralat testis (ml)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>29</td>
<td>7</td>
<td>left</td>
<td>left</td>
<td>-</td>
<td>2.0</td>
<td>m+p+s</td>
<td>+</td>
<td>12.4</td>
<td>13.5</td>
</tr>
<tr>
<td>2</td>
<td>24</td>
<td>8</td>
<td>left</td>
<td>left</td>
<td>-</td>
<td>2.0</td>
<td>p+s</td>
<td>-</td>
<td>6.2</td>
<td>9.3</td>
</tr>
<tr>
<td>3</td>
<td>26</td>
<td>10</td>
<td>left</td>
<td>left</td>
<td>-</td>
<td>2.7</td>
<td>m+p+s</td>
<td>+</td>
<td>6.9</td>
<td>20.3</td>
</tr>
<tr>
<td>4</td>
<td>24</td>
<td>7</td>
<td>left</td>
<td>left</td>
<td>-</td>
<td>2.0</td>
<td>p+s</td>
<td>+</td>
<td>8.9</td>
<td>22.1</td>
</tr>
<tr>
<td>5</td>
<td>28</td>
<td>8</td>
<td>bilat.</td>
<td>left</td>
<td>mild pain</td>
<td>1.8</td>
<td>m</td>
<td>-</td>
<td>9.7</td>
<td>12.9</td>
</tr>
<tr>
<td>6</td>
<td>27</td>
<td>5</td>
<td>left</td>
<td>left</td>
<td>moderate pain</td>
<td>2.3</td>
<td>s</td>
<td>-</td>
<td>3.7</td>
<td>10.0</td>
</tr>
<tr>
<td>7</td>
<td>30</td>
<td>10</td>
<td>left</td>
<td>left</td>
<td>moderate pain</td>
<td>2.0</td>
<td>m+p</td>
<td>+</td>
<td>7.7</td>
<td>10.4</td>
</tr>
<tr>
<td>8</td>
<td>30</td>
<td>4</td>
<td>bilat.</td>
<td>left</td>
<td>-</td>
<td>1.9</td>
<td>S</td>
<td>+</td>
<td>8.8</td>
<td>20.4</td>
</tr>
<tr>
<td>9</td>
<td>23</td>
<td>5</td>
<td>left</td>
<td>left</td>
<td>-</td>
<td>1.6</td>
<td>m+p</td>
<td>+</td>
<td>7.5</td>
<td>17.2</td>
</tr>
</tbody>
</table>

ORP = orchidopexy, dm = diameter, m = mediastinum, p = parenchyma, s = subcapsular
Figure 1 Colour Doppler ultrasound images of a left testis with a dilated intratesticular vein (diameter 3-4 mm) extending from the mediastinum to the periphery, showing absent flow during rest (left image) and retrograde flow (white arrow) during Valsalva manoeuvre (right image). Findings are consistent with an intratesticular varicocele. Note an extratesticular varicocele is present as well.
Figure 2 Colour Doppler ultrasound images of a left testis with multiple serpentine veins located mediastinal and subcapsular, showing absent flow during rest (left image) and retrograde flow during Valsalva manoeuvre (right image). Findings are in keeping with an intratesticular varicocele.
The mean age at orchidopexy of the 9 patients with ITV (8.9 years, SD ± 2.9, range 5.2–12.6) were comparable with the patients without ITV (9.5 years, SD ± 2.7, range 2.4–13.9 years; p = 0.463). Furthermore, at follow-up no significant age difference was found between the group with ITV (mean age 27.7 years, SD ± 2.7, range 22.8–30.9 years) and without ITV (mean age 26.0 years, SD ± 3.1, range 14.0–31.6 years; p = 0.110).

In all 9 patients, the ITV was found in the left, orchidopexied testis. Two of these 9 patients had undergone bilateral orchidopexy. Two patients reported occasional testicular pain, 1 of them had a history of epididymitis. None of the 9 patients with ITV had a wish for fatherhood at the time of the follow-up.

The localization of ITV was evenly distributed among the subcapsular (n=6), the parenchyma (n=6) and the mediastinum (n=5). An ipsilateral ETV was found in 6 cases, whereas none of the patients had a contralateral ETV. Besides, none of the 105 participants was found to have ETV without ITV.

The volumes of the 9 testes with ITV ranged from 3.67–12.35 ml (mean 7.96 ml, SD ± 2.42). This was significantly smaller than the volumes of the 128 orchidopexied testes without ITV (mean volume 10.45 ml, SD ± 3.46, range 2.75–20.40 ml; p=0.026, see Table 2).

Table 2 Testicular volumes as measured in the long-term follow-up study on testicular position and volume after prepubertal orchidopexy for acquired undescended testis; orchidopexied acquired undescended testis (n = 137) compared with the contralateral part (n = 73). The orchidopexied testes are subdivided in testes with (n = 9) and without (n = 128) ITV.

<table>
<thead>
<tr>
<th>Testis volume (ml)</th>
<th>N</th>
<th>mean</th>
<th>standard deviation</th>
<th>min</th>
<th>max</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Contralateral testis</td>
<td>73</td>
<td>14.11</td>
<td>4.23</td>
<td>4.80</td>
<td>23.87</td>
<td></td>
</tr>
<tr>
<td>Orchidopexied testis</td>
<td>137</td>
<td>10.28</td>
<td>3.45</td>
<td>2.75</td>
<td>20.40</td>
<td>0.000*</td>
</tr>
<tr>
<td>without ITV</td>
<td>128</td>
<td>10.45</td>
<td>3.46</td>
<td>2.75</td>
<td>20.40</td>
<td></td>
</tr>
<tr>
<td>with ITV</td>
<td>9</td>
<td>7.96</td>
<td>2.42</td>
<td>3.67</td>
<td>12.35</td>
<td>0.026**</td>
</tr>
</tbody>
</table>

* independent t-test
** Mann-Whitney U test
**DISCUSSION**

A remarkably high ITV prevalence of 8.6% was found in a long-term follow-up study of patients after prepubertal orchidopexy for acquired undescended testis. All ITVs were found in the operated testes and all were left-sided. Furthermore, our study shows that orchidopexied testes with ITV are smaller than orchidopexied testes without ITV. Das et al ⁶, Kessler et al ⁷ and Tétreau et al ⁸ reported a prevalence of 1.7%, 2.0% and 0.4%, respectively, in cohorts of men examined with scrotal ultrasound for various testicular problems, including pain, swelling, ETV, hernia and trauma. Das et al ⁶ reported on a cohort of 1,040 men, of whom 31 patients were referred for scrotal ultrasound with the diagnosis of infertility. Five (16%) of these were found to have an ITV. Bucci et al ⁹ described a cohort of 342 men who received scrotal ultrasound for routine andrological evaluation; 7 men were found to have an ITV (2.0%), and 5 of these had a history of infertility.

In the literature on ITV, a total of 74 patients with an ITV are described, mainly in case-reports.², ³, ⁶–⁹, ¹¹–²⁷ It becomes clear from the literature that only a minority of the patients seem to have any kind of symptoms. Pain (35.1%), epididymitis (5.4%), and swelling (9.4%) have been reported. This is comparable with the results of our study. In the literature, 18 patients (2.4%) were reported as infertile or as having a history of infertility. Similar to our findings, in 62% of the described cases ITV and ETV appear combined. Of the 74 reported cases, 15 (2%) had a history of inguinal surgery (8 for undescended testis, 3 for inguinal hernia, 1 for hydrocele and 3 for unknown reasons). The correlation between a history of inguinal surgery and ITV has been mentioned by several authors. Tétreau et al ⁸ described 8 cases of ITV in a cohort of 1,832 men, 5 of whom had a history of scrotal surgery, including undescended testis (n=4) and hydrocele (n=1); Zampieri et al ⁴ stated that the prevalence of ETV is increased in patients who had undergone inguinal or scrotal surgery. The high prevalence of ITV in our study shows that ITV also seems to be related to a history of scrotal and inguinal surgery.

In the present follow-up study, all ITV were found in the left, orchidopexied testis. In the literature, most of the 74 reported cases of ITV are unilateral on the left side (67.6%), 10 (13.5%) are unilateral on the right side and 14 (18.9%) are bilateral. This left-side predilection for ITV as well as for ETV is a known phenomenon which may be
explained by the fact that the left testicular vein is longer and enters the left renal vein at a right angle. This may reduce the venous return and result in an elevated residual pressure in the spermatic veins. Another theory is that the left testicular artery arches over and compresses the testicular vein. Moreover, if the sigmoid colon is distended with faeces, it can compress the vein and thus cause elevated pressure.\textsuperscript{19}

To the best of our knowledge, this is the first study on the prevalence of ITV in a cohort of men after orchidopexy. A high prevalence of ITV was found in the testes orchidopexied for acquired undescended testis. This finding leads to the question whether it is the orchidopexy or the acquired undescended testis itself which forms a risk factor for developing an ITV. It is difficult to give a definitive answer to this question. Even though every care is taken to prevent this, it might be possible that the testicular vessels are damaged during mobilization of the testis, which may result in incompetent valves of the pampiniforme plexus and dilatation of the intratesticular veins. To give a thorough answer to this question, the prevalence of ITV in the testes orchidopexied for acquired undescended testis should be compared with the prevalence of ITV in the spontaneously descended acquired undescended testes.

Previous research has shown that the volume of the acquired undescended testis after orchidopexy is smaller than both the contralateral, non-operated testis and normative values.\textsuperscript{10} In the present study, it was found that the orchidopexied testes with ITV were even smaller than the orchidopexied testes without ITV ($p = 0.026$). Smaller testicular volumes with ITV have been described before.\textsuperscript{2, 3, 7, 8, 11, 14, 15, 11, 21, 22} ITV might be the reason for a smaller testicular volume. Alternatively, the development of ITV may be a consequence of testicular atrophy as a result of a deterioration of the properties of testicular tissue, which allows enlargement of intratesticular venous structures. Besides, the mean age at orchidopexy was 9.2 years and it might be possible that some of the boys had some degree of atrophy of their undescended testis at the time of operation due to the non-scrotal position.

Unfortunately, the final explanation of the high prevalence of ITV in our post-orchidopexy study cohort remains unknown and should be a subject for further research.

Limitations of this study need to be addressed. Regarding the study design, the prevalence is based on the sonographic examination performed in the study on the long-term position and growth of the acquired undescended testis after prepubertal orchidopexy. This examination did not include colour Doppler ultrasound and although
the Valsalva manoeuvre was performed to assess signs of herniation or ETV, the testis was not specifically screened for ITV. If ITV was suspected, the patient was invited for another scrotal sonography, including colour Doppler, to diagnose ITV and to collect the data for the present study. This limitation in study design may have resulted in an underestimation of the prevalence of ITV.

CONCLUSION

A remarkably high ITV prevalence of 8.6% was found in a cohort of men in whom scrotal ultrasound was performed after prepubertal orchidopexy for acquired undescended testis. Furthermore, testicular volumes in patients with ITV were found to be smaller than in patients without ITV.
References