CASE REPORT

Transverse testicular ectopia associated with hemiscrotal hemangioma

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KEYWORDS

capillary hemangioma; transverse testicular ectopia; trans-septal orchiopexy

Summary

Transverse testicular ectopia is a rare congenital disorder characterized by both testes descending from the same inguinal canal. We report three cases of variant presentations of transverse testicular ectopia: no inguinal hernia, both testes in one hemiscrotum, and capillary hemangioma in the contralateral hemiscrotum. All patients received a simple trans-septal orchiopexy. Follow-up revealed no inguinal hernia or testicular atrophy.

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1. Introduction

Transverse testicular ectopia (TTE), also known as testicular pseudoduplication, unilateral double testes, transverse aberrant testicular maldescent, and crossed testicular ectopia, is a rare form of testicular ectopia with an unclear etiology.1–3 In patients with this disorder, both testes migrate towards the same hemiscrotum, leaving an empty hemiscrotum on the other side. Patients reported in the literature typically presented with uniform symptoms of an inguinal hernia on one side and an impalpable testis on the other side, and most patients underwent operations prior to receiving a definite diagnosis.1–4 This report describes three cases of TTE with variant presentations.

2. Case report

In the past 3 years, three boys, aged 6 months, 7 months, and 6 years, were brought to our pediatric surgery clinic with impalpable testes found since birth on the left, left, and right sides, respectively. No family history of the disorder or remarkable birth history was noted in any of the patients. Physical examination revealed two testes in the contralateral hemiscrotum and the absence of a testis on one side. Figs. 1–3 show capillary hemangiomas present on the surface of the empty hypoplastic hemiscrotum in all patients. Abdominal computed tomography showed no evidence of urinary system abnormalities or rudimentary Müllerian duct structures (uterus, fallopian tube, and ovary). The operative procedure included trans-septal delivery of the more distal testis to the empty side and
orchiopexy in the subdartos pouch (Fig. 4). One-year follow-up revealed no testicular atrophy, hernia, or resolution of the hemangiomas in all patients.

3. Discussion

TTE was first described by Von Lenhossek in 1886 in an autopsy case; since then, >100 cases have been reported. Familial occurrences have been reported, and most were associated with persistent Müllerian duct syndrome. Typically, patients present with uniform symptoms of an inguinal hernia on one side and an impalpable testis on the other. The ectopic testes were usually located in deep inguinal rings and were not detected prior to surgery. Each testis has a corresponding spermatic cord, but in most cases, the two cords fuse to form an inseparable thick-walled structure that is several centimeters proximal to the testes. No report on the association of scrotal hemangioma with this disease exists. However, in all three cases of TTE that we encountered, capillary hemangiomas were present on the empty hemiscrota. Moreover, no symptoms of inguinal hernias were present, and the ectopic testes had descended completely into the contralateral hemiscrotum.

The pathological anatomy of TTE has been elucidated by laparoscopic findings. Each testis has its own blood supply from the appropriate side. When passing into the internal inguinal ring, the spermatic vessels of the ectopic testis cross the midline and collide with the contralateral internal ring, along with the spermatic cord of the other side. Both testes usually share a single patent processus vaginalis. Fusion of the two spermatic cords in the inguinal canal is common and is usually inseparable. Several theories have been proposed to explain this maldescent, but they are inconclusive. These include obstruction of the internal inguinal ring, persistent Müllerian duct structures (hindering testicular descent), fusion of Wolffian ducts, and defective ipsilateral gubernacular development.

Testicular ectopia is associated with genitourinary tract abnormalities including horseshoe kidney, seminal vesicle cyst, persistent Müllerian duct syndrome, hypospadias, and intersex. Malignant transformation (embryonal carcinoma, seminoma, yolk sac tumor, teratoma, and mixed...
germ cell tumors) has been reported in 5–18% of patients with TTE.\textsuperscript{11}
TTE has seldom been diagnosed preoperatively because ectopic testes are usually not identified during physical examination. Fortunately, after exploration of the inguinal canal and dissection of the hernial sac, the ectopic testis can be found in the deep inguinal ring; a correct diagnosis can then be established, and both herniorrhaphy and orchiopexy can be performed without difficulty in most cases. A modified Ombredanne operation has been recommended to bring down both testes through the same inguinal canal and place each testis on its respective side of the scrotum without dividing the fused spermatic vessels and vasa deferens.\textsuperscript{5–7} Another surgical modality is extraperitoneal transposition of the ectopic testis cephalad to the penile root,\textsuperscript{3,4} but this procedure requires separating the fused spermatic cords and is not feasible in every case. Several authors have recommended using laparoscopy to elucidate the anatomy of ectopic gonads and identify Müllerian duct remnants.\textsuperscript{6,8} To obtain a precise preoperative diagnosis and to enable localizing an impalpable testis, ultrasonography, computed tomography, magnetic resonance imaging, and even arteriography and venography have been used.\textsuperscript{6–9}

We report three cases of TTE with variant presentations: both testes in one hemiscrotum, no inguinal hernia, and capillary hemangioma on the empty hemiscrotum. Since there was no inguinal hernia and both testes were in the hemiscrotum, a simple trans-septal orchiopexy was the only procedure, avoiding unnecessary inguinal exploration. Follow-up revealed no inguinal hernia or testicular atrophy. As far as we are aware, association of capillary hemangioma with TTE has not been reported in the literature, but hemangiomas were present in all three of our cases of TTE; this suggests that the occurrence is not a coincidence, although the mechanism is not yet fully understood. Further follow-up is required to identify hernias, hemangiomas, and possible malignant transformations in the testes.

References