Successful Treatment of Transverse Testicular Ectopia by Laparoscopically Assisted Orchiopexy

Eri TEI[*1], Hitoshi HIRAKAWA[*1], Masaharu MORI[*2], Naoko KOMATSUZAKI[*2], Tomoshige UMEYAMA[*2], Takaya HORIE[*2], Ryoutaro OHIZUMI[*2], Kouji IDENAWA[*2] and Toshihiko WATANABE[*2]

[*1]*Department of Pediatric Surgery, Tokai University Hachioji Hospital*
[*2]*Department of Pediatric Surgery, Tokai University School of medicine*

(Received November 15, 2023; Accepted December 28, 2023)

Transverse testicular ectopia (TTE) is a rare congenital malformation where both testes descend through the same inguinal canal and are located in the same hemiscrotum. It is usually treated with transseptal orchiopexy. In this article, we report the case of a 1-year-old boy diagnosed with TTE who was successfully treated with laparoscopically assisted orchiopexy by going through the anatomical conventional route.
A four-month-old boy was referred to our department with bilateral empty scrotum. On the physical examination, the left testis was palpable in the left groin region and the right testis was impalpable. A follow up ultrasonography was performed after 4 months, and an oval-shaped testis-like structure was detected in left internal inguinal ring near the left testis. Right side TTE was suspected in the initial diagnosis. Laparoscopic surgery was performed at age one. The left testis was observed in the inguinal canal, and the right testis was ectopically located in the left opening inguinal canal above the left testis. Two spermatic cord and testes were separated respectively, and the right testis was pulled into abdominal space laparoscopically and brought down to the right hemiscrotum via the right inguinoscrotal canal. Bilateral orchiopexy was performed via the normal anatomical route.
The postoperative course was uneventful, and testes were in the scrotum bilaterally one year after orchiopexy.

Key words: Transverse testicular ectopia, laparoscopically assisted orchiopexy, transseptal orchiopexy

INTRODUCTION

Transverse testicular ectopia (TTE) is a rare congenital malformation where both testes descend through the same inguinal canal and located in the same hemiscrotum. It is classified into three groups [1]. Type 1 is simple TTE associated with inguinal hernia (40−50%). Type 2 is TTE associated with persistent Müllerian remnants (30%) and Type 3 is associated with other anomalies such as hypospadias (20%) [2]. The patients usually present with contralateral nonpalpable testis or accidentally diagnosed intraoperatively [3]. Since it is often difficult to separate the bilateral spermatic cord, usually treated with transseptal orchiopexy. However, the laparoscopically assisted orchiopexy, which follows the original descending route of the testis, is the most ideal technique because it fixes the testis in a more physiological way and avoids bilateral orchidectomy in the event of tumor development.

Here, we present a case report that has a successful treatment of transverse testicular ectopia by laparoscopically assisted orchiopexy via the normal anatomical route.

CASE REPORT

A four-month-old boy was referred to our department with bilateral empty scrotum. He had no past medical history. On the physical examination, the left testis was palpable in the left groin region and the right testis was impalpable. In ultrasonography, the left testis was detected in the inguinal canal with inguinal hernia, but the right testis was not detected at this moment. The follow up ultrasonography was performed after 4 months, and an oval-shaped testis-like structure measuring 9 x 5 mm in size (Fig. 1) was detected in left internal inguinal ring near the left testis. The size of the left testis was 11 x 7 mm. Right side TTE was suspected in the initial diagnosis. There was no evidence of Müllerian duct structures. Laparoscopic surgery was performed by using a 5 mm telescope from the umbilicus at age one. The left testis was observed in the inguinal canal and the right testis was ectopically located in the left opening inguinal canal above the left testis (Fig. 2). Patent processus vaginalis (PPV) was not observed on the right side (Fig. 3). Each testis had a separate spermatic cord. There was no evidence of Müllerian duct structures such as small uterus. We opened the inguinal canal by using the standard technique of herniotomy. Two spermatic cords and testes were separated respectively, and high ligation of the hernia sac was performed (Fig. 4). As adequately detached spermatic cords were observed laparoscopically, intraperitoneal spermatic cord separation was not necessary. A route from just beside the right internal

Eri TEI, Department of Pediatric Surgery, Tokai University Hachioji Hospital, 1838 Ishikawa-machi, Hachioji, Tokyo 192-0032, Japan
Tel: +81-42-639-1111 Fax: +81-42-639-1112 E-mail: e-tei@tsc.u-tokai.ac.jp

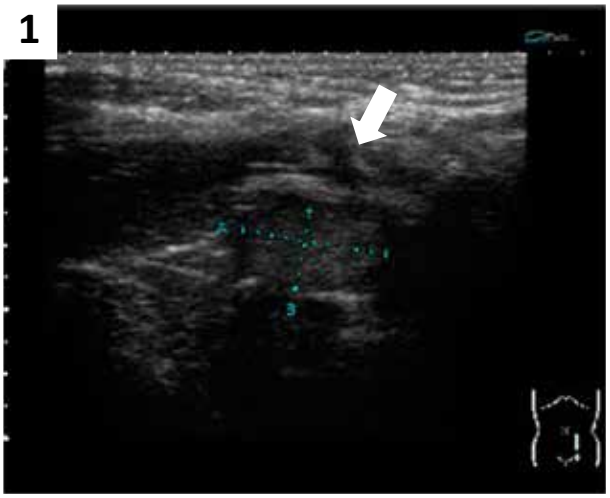

Fig. 1 Ultrasound revealing the oval-shaped testis-like structure, measuring 9 x 5 mm in size (white arrow).

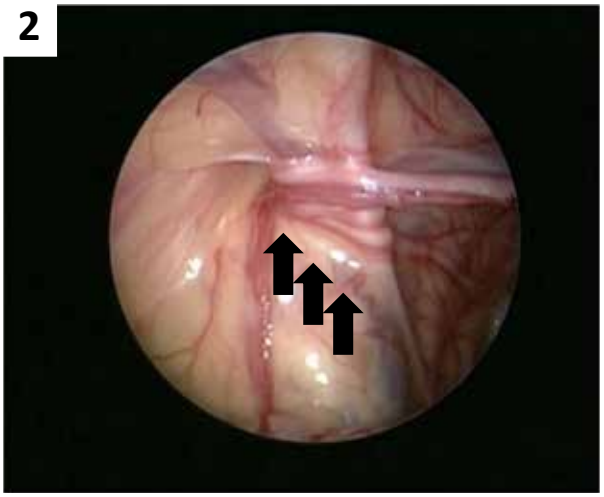

Fig. 2 Laparoscopic image showing the right testis entering the left internal inguinal ring along with the left testis (black arrow).

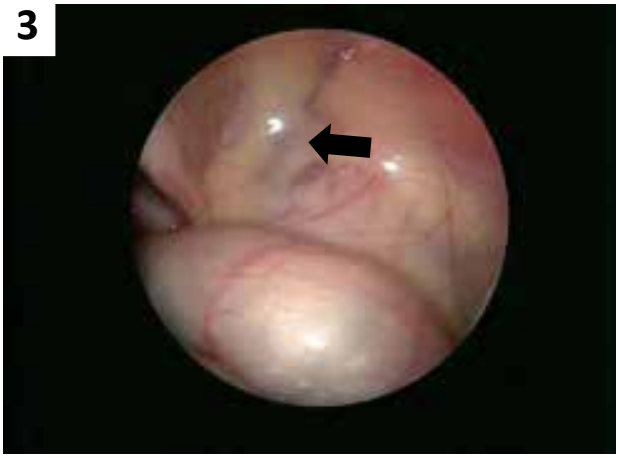

Fig. 3 Patent processus vaginalis (PPV) was not observed on the right side (black arrow).

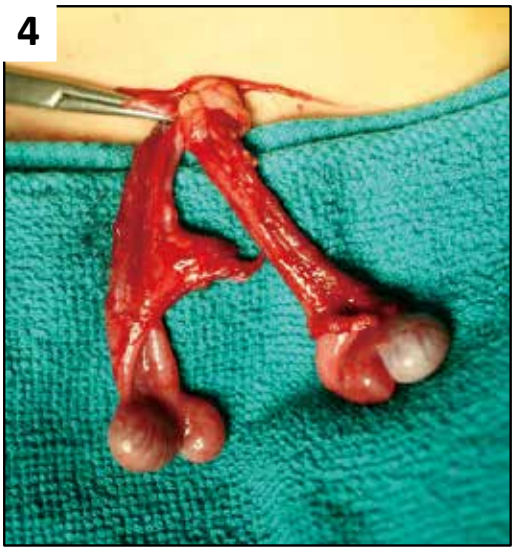

Fig. 4 Two spermatic cord and testes were separated respectively.

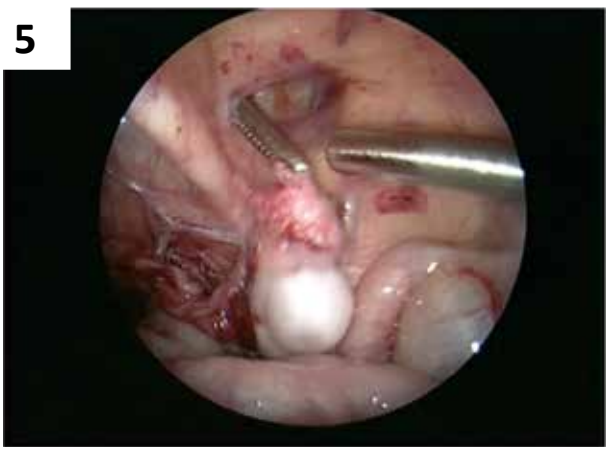

Fig. 5 Pulled the right testis into abdominal space laparoscopically.

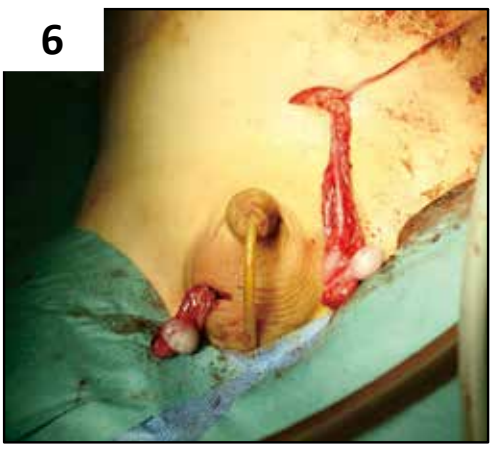

Fig. 6 The right testis brought down to the right hemiscrotum via the right inguinoscrotal canal.

ring to the right hemiscrotum was created with Kelly forceps. The right testis was pulled into abdominal space laparoscopically (Fig. 5) and brought down to the right hemiscrotum via the right inguinoscrotal canal (Fig. 6). Bilateral orchiopexy was performed via the normal anatomical route. The defective peritoneum was sutured as much as possible.

The postoperative course was uneventful, and testes were in the scrotum bilaterally, one year after orchiopexy. Three years after the surgery, no late complications such as testicular atrophy, re-elevation, or tumor development have been observed.

DISCUSSION

TTE is a rare condition in which one testis crosses the median into the inguinal canal of the opposite testis and may be discovered incidentally during surgery for inguinal hernia or nonpalpable testis. The first case was reported by Von Lenhossek in 1886 [4]. Although more than 100 cases of TTE have been reported in the English literature, the exact frequency of occurrence is still remains unknown. There are several theories as to the cause, including Wolffian duct fusion, Müllerian remnants, and atrophy, contracture, or abnormal attachment of the gubernaculum testis [5–6], but these have not yet been elucidated.

As I mentioned above, there are three types of TTE [7]. Our case is type 1 which is the most frequently observed. Laparoscopy is often used to diagnose TTE and observe the type of TTE but is rarely used as an actual surgical approach. Most cases have been reported with transseptal orchiopexy due to its minimally invasive approach. Since the development of malignancies were reported as a serious complication [8–10], the laparoscopically assisted orchiopexy, which follows the original descending route of the testis, is considered with the most ideal technique. There have been reports of embryonal carcinoma, seminoma, yolk sac tumor, and teratoma [11]. Fixation of bilateral testes by the original route could avoid bilateral orchidectomy after development of malignancies. In our case, the spermatic cord was safely separated without adhesions and the spermatic cord of the TTE was long enough to secure the testis in the scrotum by the original inguinal canal route.

In the case of type 2, preoperative diagnosis of persistent Müllerian duct syndrome (PMDS) can be made as a result of a preoperative magnetic resonance imaging (MRI) scan that confirms the presence of fallopian tubes, a uterus and an upper part of vagina. However, in the most cases Müllerian remnants were recognized with laparoscopic investigation. Müllerian remnants are extremely hard to split in half, and there should be an option of transseptal orchiopexy or bilateral orchidectomy to avoid future malignant transformation. The occurrence of malignancies such as adenocarcinoma, cystadenocarcinoma, and squamous cell carcinoma after puberty has been reported in previous studies [8–10]. The main purpose of treatment of TTE is to preserve fertility. The second purpose is to prevent future malignant transformation. Fertility of PMDS is rarely reported [12]. Surgical excision of the infantile uterus and fallopian tubes risks damage to vasa spermatic cord and the deferential blood supply to the testis. Although malignant transformation of PMDS is reported to be around 18% [8–10], it is desirable to preserve the testes as much as possible to preserve fertility, and long-term follow-up should be essential when Müllerian remnants are preserved. Type 3 cases can be treated by Type 1 surgical procedures, apart from treating complications.

Orchiopexy should be performed after six months and before 18 months of age to reduce the risks of infertility and development of cancer [13]. As the most desirable treatment for TTE, first it is necessary to observe laparoscopically whether both spermatic cords can be separated and, if so, whether there is enough distance to fix the TTE in the original scrotum. Secondly, the TTE should be separated intraperitoneally, and the testis fixed through the original inguinal canal, but if this is not possible, both testes should be withdrawn from one inguinal canal and performed transseptal orchiopexy. The occurrence of malignancies from Müllerian remnants has been reported, and resection of the remnant tissue is originally recommended [8–10]. However, the majority of cases will probably leave the Müllerian remnants because of the risk of damaging the spermatic cord during the operation.

We present a case of successful laparoscopic treatment of TTE via the normal anatomical route. The laparoscopically assisted orchiopexy has been rarely performed due to the difficulty of the surgical technique. However, the development of laparoscopic techniques has made this surgery possible. The laparoscopic surgery allows safe separation of testes with early recovery and excellent aesthetic outcomes. Moreover, this method fixes the testis in a more physiological way and avoids bilateral orchidectomy in the event of tumor development. However, indication needs to be carefully considered with each case.

DISCLOSURE

The authors declare no conflicts of interest.

AUTHOR CONTRIBUTIONS

E.T. wrote the manuscript. H.H. contributed to the conception and revised the manuscript. H.H. is the primary assistant during the surgery. All authors read and approved the final manuscript.

HUMAN ETHICS

This case report is for academic communication only and not for other purposes. The patient provided informed consent, and patient anonymity was preserved. The approval of our institutional ethics committee was unnecessary for a clinical case report.

REFERENCES

1) Esteves E, Pinus J, Maranhao RF, Abib S de C, Pinus J. Crossed testicular ectopia. Sao Paulo Med J 1995; 113(4): 935–40.
2) Gauderer MW, Grisoni ER, Stellato TA, Ponsky JL, Izant Jr RJ. Transverse testicular ectopia. J Pediatr Surg 1982; 17(1): 43–7.
3) Bascuna R Jr, Ha JY, Lee YS, Lee HY, Im YJ, Han SW. Transverse testis ectopia: diagnostic and management algorithm. Int J Urol. 2015; 22: 330–331.
4) Von Lenhossek MN. Ectopia testis transversa. Anat Anz. 1886; 1: 376–381.
5) Josso N, Belville C, di Clemente N, Picard J-Y. AMH and AMH receptor defects in persistent Müllerian duct syndrome. *Human Reproduction Update*. 2005; 11(4): 351–356.

6) Gujar N.N. Male form of Persistent Mullein Duct Syndrome Type 1 (hernia uteri inguinalis) presenting as an obstructed inguinal hernia: a case report. J Med Case Rep. 2011; 5: 586.
7) Yıldız A, Yiğiter M, Oral A, Bakan V. Transverse testicular ectopia. Pediatr Int. 2014; 56: 102–105.
8) Romero FR, Fucs M, Castro MG, Garcia CR, Fernandes Rde C, Perez MD. Adenocarcinoma of persistent Müllerian duct remnants: case report and differential diagnosis. Urology 2005; 66: 194–5.
9) Shinmura Y., Yokoi T., Tsutsui Y. A case of clear cell adenocarcinoma of the müllerian duct in persistent müllerian duct syndrome: The first reported case. Am. J. Surg. Pathol. 2002; 26: 1231–1234.
10) Thiel DD, Erhard MJ. Uterine adenosarcoma in a boy with persistent müllerian duct syndrome: first reported case. J Pediatr Surg. 2005; 40: e29–31.
11) Zhu Y P, Zhang S L, Ye D W, Shi G H, Xiao W J. Yolk sac tumor in a patient with transverse testicular ectopia. World J Surg Oncol. 2011; 9: 91.
12) Viart L, Peltier J, Forzini T, Page C, Foulon P, Saint F, *et al*. Persistent Müllerian ducts syndrome: one case of late hypofertility. Morphologie. 2015; 99: 23–28.
13) Chan E, Wayne C, Nasr A., FRCSC for Canadian Association of Pediatric Surgeon Evidence-Based Resource. Ideal timing of orchiopexy: a systematic review. Pediatr Surg Int. 2014; 30(1): 87–97.