

CASE REPORT

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A case of abdominal migration of the testes presenting with azoospermia

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ABSTRACT

31-year-old male was referred to our hospital due to azoospermia. Physical examination revealed impalpable testes in the scrotum, and operative scar presented at lower abdominal midline. Magnetic resonance imaging (MRI) revealed that small testes were located subcutaneously in the lower abdominal midline. Since luteinizing hormone (LH) and follicle stimulating hormone (FSH) were elevated, we diagnosed non-obstructive azoospermia (NOA) due to abdominal migration of the testes. Microscopic testicular sperm extraction (micro-TESE) was performed, however, no sperm were recovered. Pathological diagnosis was Sertoli cell only and no malignant cells were observed. Post-operatively, subjects' hormone levels were unchanged, and testicular tumor markers and computed tomography (CT) were normal. However, renal function gradually deteriorated and a renal transplantation from the farther was carried out eight months after micro-TESE. Attention to the possibility of carcinogenesis of the abdominal migrated testes should be maintained.

Keywords: abdominal migration of testes, NOA, micro-TESE

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INTRODUCTION

Among cases of cryptorchidism, impalpable testes have been reported to be about 20%.¹ Impalpable testes among adults is a risk factor for testicular cancer. We report here on the case of a 31-year-old male, who wanted to be a father, with abdominal migration of the testes and presenting with non-obstructive azoospermia (NOA).

CASE REPORT

31-year-old male attended our hospital due to azoospermia. He was 170cm tall, 59Kg body weight, and body mass index (BMI) was 20.8. Although he had a history of surgery for cryptorchidism as an infant, physical examination revealed midline surgical wound in the lower abdominal region and lack of scrotal contents, which he had previously noticed. He had a history

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of membranous nephritis, and his creatinine value ranged from 3.2 to 4.0 and required a protein diet by physician. Semen analysis revealed azoospermia. Endocrine panel showed high gonadotropin values (luteinizing hormone=LH:13.9 mIU/mL, follicle stimulating hormone=FSH:33.8 mIU/mL), whereas testosterone was 6.956 ng/mL. No serum tumor markers indicating testicular cancer were elevated. Chromosomal G banding was 46XY and no Y chromosomal microdeletion observed. Abdominal MRI showed that testes were of tiny mass and located subcutaneously in the lower abdominal midline. (Figure-1) No adrenal tumor was detected. We diagnosed NOA with abdominal migration of the testes.

Under epidural anesthesia, micro-TESE was performed along the previous lower abdominal midline skin incision. The testes were firmly adhered subcutaneously and finally isolated according to the MRI finding. (Figure-2) However, testicular blood vessels were not clearly identified. Testes were subjected to horizontal vertical incision for micro-TESE procedure. Under a surgical microscope, only atrophic seminal tracts with rich stroma were observed. No sperm was detected from retrieved tissue bilaterally. Pathological diagnosis was Sertoli cell only and no malignant cells were observed. The patient and his wife desired donor insemination (DI) and were referred for treatment. Fortunately, his wife conceived at initial attempt of DI, and twin healthy girls were born.

The patient was followed by endocrine panel, tumor markers, and CT to exclude malignant changes to the abdominal migrated testes. Although hormone, tumor markers (Table 1), and CT were within normal range, his renal function gradually deteriorated. Eight months later after micro-TESE, he received a renal transplantation from his father and his creatinine level recovered.

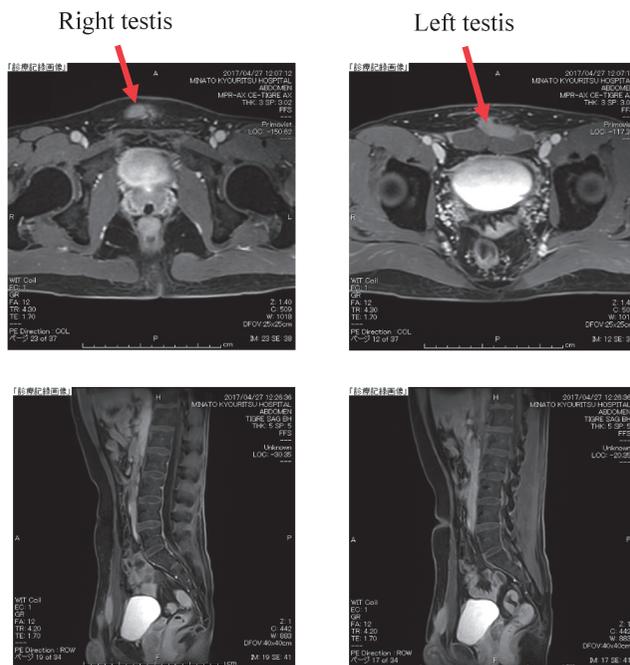


Fig. 1 MRI revealed that right testis existed at almost center of lower abdomen and left testis located above the abdominal rectal muscle subcutaneously

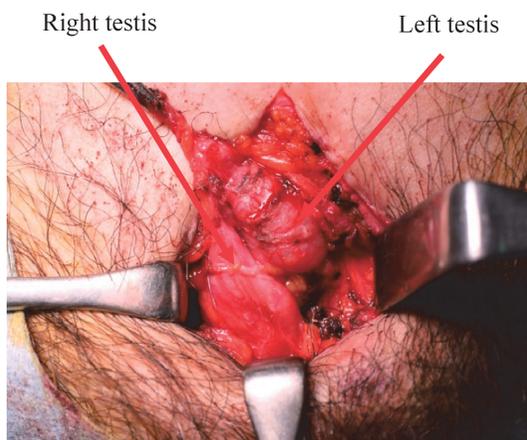


Fig. 2 Intraoperative finding

The testes existed subcutaneously just as MRI findings

Table 1 Endocrine panel and tumor markers pre- and post-surgery

	before surgery	3 months after surgery	6 months after surgery	14 months after surgery
LH mIU/mL	13.9	18.5	23.6	12.5
FSH mIU/mL	33.8	34.2	36.7	30.3
Testosterone ng/mL	6.956	8.363	9.601	8.111
Free-testosterone pg/mL	6.1	14.4	13.6	8.8
HCG mIU/mL	0.5↓	–	0.5↓	0.5↓

Decreased testosterone after micro-TESE was not observed.

DISCUSSION

Causes of NOA are genetic mutations, chromosomal aberrations, hormonal disorders, mal-descent of testis, the use of certain medications, radiation, and the presence of toxins. NOA is characterized with elevated FSH and atrophic testes. TESE is now widely applied as a method of sperm retrieval surgery, and there is no doubt that micro-TESE has become a standard technique for NOA patients.²

Cryptorchidism is associated with impaired fertility and is a risk factor for testicular cancer. Amongst men who have had undescended testis, the risk of cancer is increased two to eight times, and 5 to 10% of all men with testicular cancer have a history of cryptorchidism.³ Thus, impalpable testis, especially undescended testis, is a critical problem. Whether the actual age at orchidopexy has any effect on the risk is unclear. Pettersson et al investigated the relationship between the age at operation for undescended testis and the subsequent risk of testicular cancer.⁴ In this cohort study of 16,983 men with surgical treatment of cryptorchidism, the risk of testicular cancer among men who were treated at 13 years of age or older was approximately twice that among men who underwent orchidopexy before the age of 13. According to the Swedish Cancer Registry, the cumulative incidence of testicular cancer up-to the age of 54 years in Sweden in 2004 was approximately 0.5%.⁴ However, it is unknown whether cryptorchidism and testicular

cancer have a common cause, or whether cryptorchidism is in itself a cause of testicular cancer.

The present case had the history of surgery for “cryptorchidism” at infancy, however, as for the details such as original testicular location, the surgical method remained unclear. Since no inguinal surgical scar was observed, the testes may originally have migrated to the abdominal lesion. Fortunately, no decreased testosterone was observed postoperatively even though the testes were small and his sexual function was not altered.

The first reported case of ectopic testis in the anterior abdominal wall was a 3-year-old boy.⁵ The anterior abdominal wall reported the rarest site of testicular ectopia at neonate or infant.⁶ No adult case has been reported as present case before. Punwani et al reviewed ectopic testes which were perineal area in 4 cases, transverse testicular ectopia in 11, and abdominal area in 2. They hypothesized that where testicular ectopia can explained that anomalous attachment of the gubernaculum to the anterior abdominal wall during transabdominal descent, or aberrant migration of the gubernaculum during the inguinoscrotal phase of testicular descent.⁷ The hypothesis of tails of Lockwood as traction theory (Charles Barrett Lockwood, 1888) explained the mechanism of testicular ectopia as the contraction of gubernaculum fibers spreading away from the normal descent pathway of the testes. However, later studies by other authors proved that the gubernaculum is initially attached to the scrotum with no fibers spreading in multiple directions.⁸ Although the cause of ectopic testes was unclear, Nightingale et al abandoned the theory of Lockwood which had been existed from more than 100 years ago.⁸

CONCLUSION

Abdominal migrated testes in adult are considered to be rare. Unfortunately, no sperm recovery was achieved, great attention to the possibility of carcinogenesis should be maintained.

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STATEMENT OF ETHICS

The protocol for this research project, including its use of human subjects, was approved by a suitably constituted Ethics Committee.

DISCLOSURE STATEMENT

Human rights and informed consent statements: All procedures completed were done in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national), and with the Helsinki Declaration of 1964 and its later amendments. Informed consent was obtained from this patient for the purpose of inclusion in this study.

Animal rights statements: This article does not contain any studies with animal subjects performed by any of the authors.

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Each author has no COI with regard to this manuscript.

REFERENCES

- 1 Kogan S.J. Controversies Regarding Treatment of Undescended Testes. In: Poppas D.P., Retik A.B. editors. *Pediatric Urology. Atlas of Clinical Urology*. Current Medicine Group, London. 2003. p.79–95.
- 2 Schlegel PN. Testicular sperm extraction: microdissection improves sperm yield with minimal tissue excision. *Hum Reprod*. 1999;14(1):131–135. doi:10.1093/humrep/14.1.131.
- 3 Dieckmann KP, Pichlmeier U. Clinical epidemiology of testicular germ cell tumors. *World J Urol*. 2004;22(1):2–14. doi:10.1007/s00345-004-0398-8.
- 4 Pettersson A, Richiardi L, Nordenskjold A, Kaijser M, and Akre O. Age at surgery for undescended testis and risk of testicular cancer. *N Engl J Med*. 2007;356(18):1835–1841. doi:10.1056/NEJMoa067588.
- 5 Rao PL, Gupta V, Kumar V. Anterior abdominal wall-an unusual site for ectopic testis. *Pediatr Surg Int*. 2005;21(8):687–688. doi:10.1007/s00383-005-1444-4.
- 6 Siddiqui SA, Marei TI, Al-Makhaita G. Testicular ectopia in the anterior abdominal wall of a neonate: A rare site of ectopic testis. *Am J Case Rep*. 2016;17:490–493. doi:10.12659/ajcr.895765.
- 7 Punwani VV, Wong JSY, Lai CYH, China JCY, Hutson JM. Testicular ectopia: Why dose it happen and what do we do? *J Pediatr Surg*. 2017;52(11):1842–1847. doi:10.1016/j.jpedsurg.2016.12.009.
- 8 Nightingale SS, Al Shareef YR, Hutson JM. Mythical “Tails of Lockwood”. *ANZ J Surg*. 2008;78(11):999–1005. doi:10.1111/j.1445-2197.2008.04720x.