CASE REPORT

Management of bilateral nubbin testis, past and present controversies: a case report

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ABSTRACT

Introduction: Testicular nubbin is the most common case of impalpable testis found by pediatric urologists. Diagnostic and surgical management, including long-term follow-up, is still a debate, especially in cases of bilateral testicular nubbin.

Case presentation: A 5-year-old boy with bilateral impalpable testis was brought to a hospital. A diagnostic laparoscopy was performed, and both internal inguinal rings were closed with testicular blood vessels, and the vas deferens had entered the inguinal canal. We found bilateral testicular nubbin on inguinal exploration and subsequently performed only right orchidopexy because it was larger than the left testis.

Conclusion: Evidence-based medicine, comprehensive management, and consultation between doctor and patient are needed in bilateral management of testicular nubbin for a good patient outcome.

INTRODUCTION

Undescended testis is one of the most common surgical conditions in pediatric patients. The incidences are 0.8% on one-year-old male babies and 3.0% of full-term male newborns at birth, whereas 20% are determined to be clinically impalpable.1,2 One or more impalpable testicles may be found in the peritoneal cavity, inguinal canal, in an ectopic position, or as a little aberrant testicular remnant/nubbin testis.1,3 Vanishing testis syndrome (VTS) or Testicular Regression Syndrome (TRS) or Nubbin testis or also known as congenital bilateral anorchia is the most common cause of impalpable testis in children (40–60%) on inguinal exploration. Identification of a nubbin testis or blind-ending vessels within the remnants between 0 and 16%.3,4,6 Thus, what about the contralateral testis? Whether to do orchidopexy or not is still a matter of debate among experts.

We report a boy with bilateral nubbin testis and their management by employing the literature review method. This case report is in line with the SCARE guidelines 2020.7,8

CASE REPORT

A 5-year-old Sundanese boy from a low socio-economic family came to the Pediatric Surgery polyclinic because both testicles were not in the scrotum. The parents only realized this when the child was one year old. The stretched penile length was normal. Based on the result of an inguinoscrotal examination, the testis was not palpable in the scrotum or inguinal bilateral. An ultrasound (US) examination of the inguinal and lower abdomen was performed with the result that no testicular structures were found in both inguinal and ectopic areas. Through intra peritoneal US scanning, it is difficult to find
The controversy surrounding inguinal exploration and excision of these testicular remnants occurred at the time of operative intervention. The other issue is the variable incidence of viable GCs reported in different studies between 0 and 16%. The incidence of GCs in pathology examination occurred around 10% and also seminiferous tubules (SNT) at 24% in the present series.

The first red flag for the diagnosis of TRS was bilateral impalpable testes in a newborn male on inguinal examination. The American Urological Association (AUA) guidelines recommend getting a karyotype, electrolytes, and 17-hydroxyprogesterone examination to rule out a genetic female (46 XX) with severe virilization owing to Congenital Adrenal Hyperplasia (CAH). By contrast, if 46 XY karyotype, an evaluation to distinguish VTS/TRS versus bilateral abdominal testis is warranted. Hormonal studies should be considered to assess the presence of any viable testicular tissue including serum Mullerian Inhibiting Substance (sensitivity 92–100%, specificity 98%) and additional hormone testing (inhibin B, FSH, LH, and testosterone). The hCG stimulation was recommended but not to diagnose anorchia.

When the patient is younger than 12 months with high LH and FSH serum, undetectable Mullerian Inhibiting Substance and inhibin, and low testosterone condition, the hCG stimulation testing or surgical exploration are unnecessary for the diagnosis of isolated anorchia. Laparoscopic surgical exploration has proven to be a practical approach for identifying and evaluating impalpable testis. According to some experts, routine exploration of the inguinal region should only be done if laparoscopy reveals an open internal ring with normal spermatic vessels. However, this exploration is not recommended if the internal ring is closed. Others agree, believing that no further treatment is necessary if blind-ending vessels are discovered in the abdomen. Testicular prostheses, a cosmetic alternative for individuals who have lost one or both testicles, can be implanted after inguinal exploration. As a result, the implantation of a testicular
prosthesis can be done as a first inguinal procedure, and testicular remnants can be removed simultaneously.\textsuperscript{11-15}

In our case report, we did not perform preoperative hormone testing, which is a limitation of the management of this patient. We carried out an inguinal exploration because this patient had bilateral testicular nubbins, so we had to investigate the possibility of a macroscopic testicular structure. The size of the right testicular nubbins was slightly larger than the contralateral side. We continue to perform right orchiopexy, hoping that the nubbins tests will produce the hormone testosterone. Until now, we could not prove this patient’s hormone levels of FSH, LH, and testosterone because the patient’s parents refused to do hormone tests due to psychosocial problems and the possibility of further hormonal therapy and testicular prosthesis implantation.

Testicular remnant orchiectomy is often performed because of the possibility of malignancy; others did not do it because they could not find testicular tissue in testicular remnants.\textsuperscript{1,8,11} The risk of testicular cancer in the nubbins would be 0 to 1.6%, with an assumed rate of 0 - 16% of remnants containing germ cells (GCs).\textsuperscript{7} To date, only one published report of intratubular GC neoplasia in cases of the testicular remnant.\textsuperscript{3,8} Several authors concluded no correlation between histopathological examination results and the incidence of malignancies in testicular nubbins, especially intratubular GC neoplasia.\textsuperscript{5,8,11,17}

In this case report, we did not perform left orchiectomy because of the evidence-based low incidence of malignancy. We will then carry out a long-term follow-up of this patient for right testicular function and possible left testicular malignancy.

CONCLUSIONS

The management of patients with impalpable testis must be based on an evidence-based data and comprehensive evaluation so that the diagnosis and treatment can be adequately determined. The condition of bilateral testicular nubbins requires good counseling for parents and patients so that there are no developmental and psychosocial disorders.

CONFLICT OF INTEREST

No potential conflicts of interest were declared.

SOURCE OF FUNDING

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

ETHICAL CONSIDERATION

Patient/Legal guardian had received signed written informed consent regarding publication of the medical data in scientific medical journal.

AUTHOR CONTRIBUTION

All author had contributed I manuscript writing and agreed for the final version of the manuscript for publication.

REFERENCES


