Urology Case Reports 39 (2021) 101828

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Polyorchidism and testicular malignancy, what can we learn: A case report

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ARTICLE INFO

Keywords:
Polyorchidism
Seminoma
Malignancy

ABSTRACT

Introduction: Polyorchidism is a rare condition with a total number of approximately 190 cases. Malignancy was found in 6,4% of cases.
Case presentation: A 57 years old man came with a sudden and persistent painful mass in right inguinal region. The patient decided to undergo surgery with diagnosis of incarserated lateral hernia inguinal and obtained a testicular-like lump in the right inguinal canal, then the patient underwent orchiectomy. Histopathological examination revealed a soft tissue tumor with microscopic characteristic of seminoma. CT-Scan revealed metastasis to lung and liver.
Conclusion: Attention must be given to detect malignancy in polyorchidism.

1. Introduction

Polyorchidism is a rare condition due to an embryological abnormal division of the genital ridge. Triorchidism is the most common form, predominantly on the left side and located intrascrotal. These supernumerary testes (SNT) are at increased risk to develop malignancy and torsion.[1]

2. Case report

A 57 years old man was referred to our emergency unit due to a sudden and persistent painful mass in right inguinal region. There was no history of fever, nausea, vomit, trauma, urinary stone, and hematuria.

Physical examination revealed a tender mass in the right inguinal area, the patient had no fever. The scrotum showed normal two testis in place.

The patient decided to undergo surgery with diagnosis of incarserated lateral hernia inguinal and obtained a testicular-like lump in the right inguinal canal, then the patient underwent orchiectomy.

Laboratorium examination revealed anemia (Hb 10,2 g/dl, Hct 34%) and thrombocytosis (Thrombocyte 665.000 cells/mm^3). Diff count test revealed decreasing of eosinophils, neutrophil bands, and lymphocytes and increasing of neutrophil segments. The patient underwent surgery and found that there was a testicle in the inguinale canalis and the type of polyorchidism is B1 (Fig. 1). The patient was examined for anatomic pathology and found an image of the germ cell carcinoma. Tested negative for CD117 and CD30 immunohistochemistry. No brown cells were found (Fig. 2).

The patient was examined for tumor markers showed lactate dehydrogenase (144 U/L), alpha-fetoprotein (1,9 IU/mL), and beta-human chorionic gonadotropin (0,19 mIU/mL) within normal limit.

Histopathological examination revealed a soft tissue tumor sized 2 × 5 cm with solid smooth mucinous lamellation with microscopic characteristic of condensed round cells proliferation with pleomorphic nucleus, with septum and connective tissues between cells suspected for germ cell tumor (differential diagnosis of seminoma and embryonal cell carcinoma). The patient was examined by CT-Scan revealing metastasis to lung and liver. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

3. Discussion

Triorchidism is the most common type of polyorchidism presented with two testes on one and one testis on the other side. In a meta-analysis performed in 2009, the median age was 17 years and most cases of

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https://doi.org/10.1016/j.eucr.2021.101828
Received 14 August 2021; Accepted 29 August 2021
Available online 30 August 2021

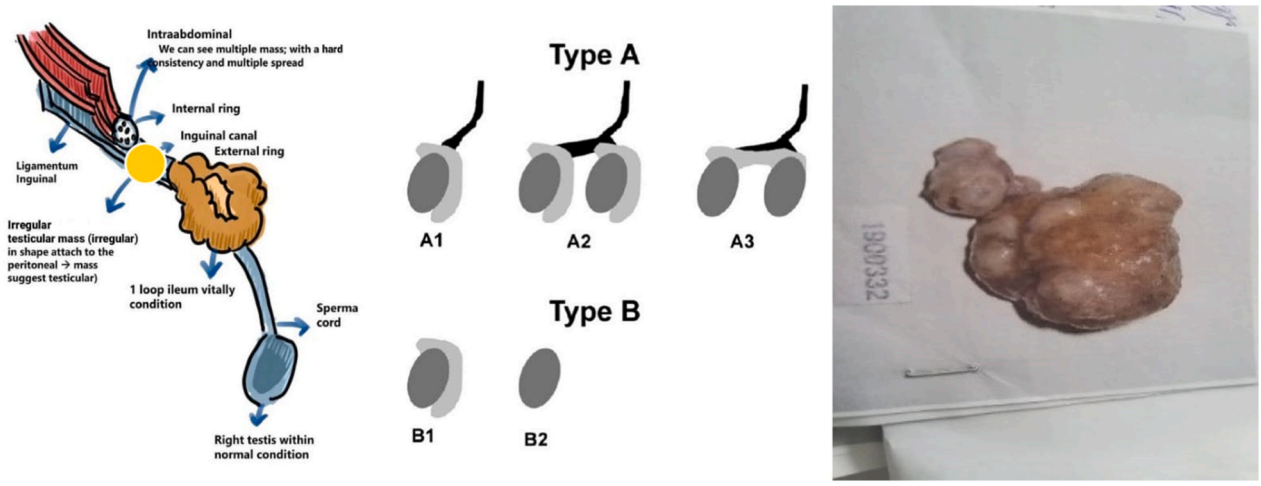

Fig. 1. A. Testis in canalis ingunale B. Type of Poliorchidism C. Tumor Size 2 × 5cm.

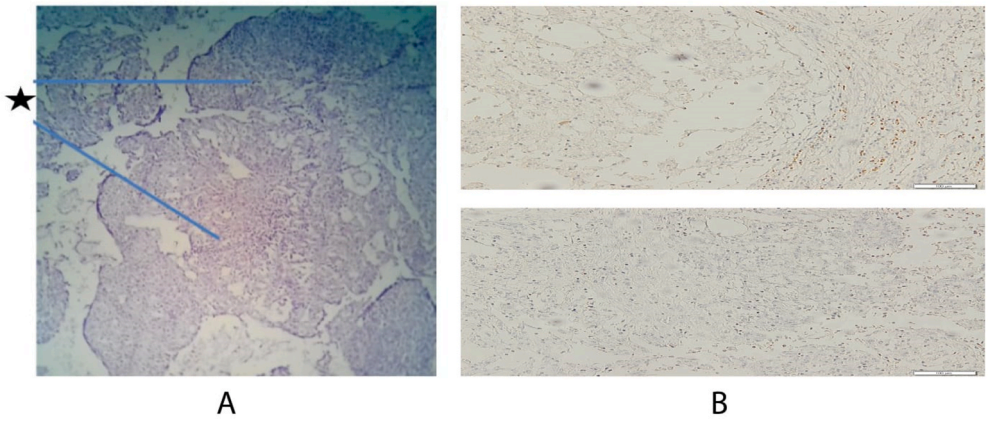

Fig. 2. A. Groups of closely packed tumour cells* B. CD117 and CD30 Immunihistochemistry not found brown cell (−). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

polyorchidism were confined to the left side (65%).[1] The most common anomalies associated with polyorchidism are inguinal hernia (30%), undescended testis (15%–30%), testicular torsion (13%) and malignancy (<1%).[2]

Most patients are asymptomatic or present with painless scrotal mass. Although it is rare, testicular cancer is the most concerning cause of a painless scrotal mass. Therefore, further evaluation with imaging work-up is warranted to exclude underlying malignancy. The estimated increased risk of malignancy in SNT is about 6%, and the commonly encountered histological type including seminoma, choriocarcinoma, and teratoma.[3] Type of malignancy can be seen in Table 1.

Embryologically polyorchidism can be classified into four types. In type A, the division separates a small part of the genital ridge, which does not contact the mesonephric duct. Therefore, the supernumerary testis lacks an epididymis and vas deferens; this type is more prone to movement and can be presented as undescended testis such as our case. In type B, the division of the genital ridge occurs in the region where the primordial gonads are attached to the mesonephric ducts and the supernumerary testis has its own epididymis. In type C, the supernumerary testis has its own epididymis and shares the vas deferens with the regular testis in a parallel fashion. In this type of polyorchidism, there is an incomplete longitudinal division of the genital ridge and the proximal portion of the mesonephric duct. In type D, which is the least common, complete longitudinal duplication of the genital ridge and mesonephric duct occurs, with resultant complete duplication of testes, epididymides, and vas deferens.[4]

Although the reported incidence of cancer is less than 1% with polyorchidism, the collective rate among some series is 6.25%–7%. According to a study by Bergholz et al., neoplasm of the SNT was found in 9 cases (6,4%). Of 9 cases, 8 were malignant (3 seminomas, 2 choriocarcinomas, 2 teratomas, 1 embryonal carcinoma) and 1 was benign (rete testis adenoma).[5]

Since it was found that 7 of 8 malignancies (88%) in non-scrotal supernumerary testis, cryptorchidism appears to be the most important risk factor for malignancy in patients with supernumerary testes. Three neoplasms (25% of all abdominal SNTs) were obtained in abdominal supernumerary testes and three neoplasms occurred in inguinal SNTs (9% of all inguinal SNTs) .[5]

Furthermore, in the general setting no urology patient is regularly screened for polyorchidism if there are 2 testes in the scrotum. This observation also may lead to a higher skewed incidence of testicular cancer in polyorchidism. However, those facts cannot completely explain the 142-fold increased incidence of malignancy in SNT compared to the general population.The management of polyorchidism is controversial. Orchiectomy is one option due to the risk of malignancy. If there is no malignancy, patients can be followed up conservatively.[5]

4. Conclusion

Currently, most malignancies due to SNT have been found at an advanced stage with metastasis. Hence, attention must be given to detect

Table 1
Case of Polyorchidism with malignancy.[5].

Author	Type	Location	Staging
Chowdary et al.	Seminoma	Scrotal	T1NxMx
Robert Bergholz et al.	Seminoma	Scrotal	N/A
Hasan Salih Sagman et al.	Seminoma	Scrotal	T1NxMx
A.H. Ahmed et al.	Choriocarcinoma	Scrotal	N/A
Robert Bergholz et al.	Teratoma	Scrotal	N/A
Oktay Ozman et al.	Teratoma	Scrotal	N/A
Robert Bergholz et al.	Embriyonal carcinoma	Scrotal	N/A
Robert Bergholz et al.	Benign Rate Testis Adenoma	Scrotal	N/A
Jaiger Chintamani et al.	Choriocarcinoma	Scrotal	T1NxMx
Robert Bergholz et al.	Seminoma	Inguinal	N/A
Robert Berg et al.	Choriocarcinoma	Inguinal	N/A
Robert Bergholz et al.	Teratoma	Inguinal	N/A
Robert Bergholz et al.	Seminoma	Abdominal	N/A
Robert Bergholz et al.	Seminoma	Abdominal	N/A
Robert Bergholz et al.	Chorocarcinoma	Abdominal	N/A
Sawkar Vijay Pramod et al.	Germ Cell Carcinoma	Inguinal	pT1NxM1bS1 (Stage IIIC)

malignancy in polyorchidism.

Sources of funding

Immanuel Hospital, Bandung, Indonesia.

Ethical approval

This manuscript was approved by Dr. Hasan Sadikin General Hospital Ethical Committee.

Informed consent

Written informed consent was obtained from the patient for publication.

Author contribution

All authors have participated sufficiently in the intellectual content, conception and design of this work and writing of the manuscript.

Guarantor

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Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

None declared.

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