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Short Communication

Testicular and Para-Testicular Tumours in Children: Clinical Aspects and Outcome Over Six Years

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Abstract

In this study, we aimed to describe clinical aspects and the outcome of testicular and para-testicular tumours in children treated in our institution.

Introduction

Testicular and para-testicular are rare in children. They represent 1% to 2 % of all solid tumours in children [1]. R. Kay [2] groups these tumours into seven groups from the germ cell to the para-testicular tumours [2]. In children, germ cell tumours are the most frequent [3]. The dosage of serum tumour markers, notably the ß-human chorionic gonadotropin (ß-HCG) and the α-fetoprotein (α-FP), is crucial for diagnosis orientation and therapeutic decision. In addition, it is essential for follow-up. Often, orchiectomy alone is sufficient for the treatment. However, for high-risk or aggressive histological forms, chemotherapy and sometimes associated radiotherapy complement the surgical treatment.

Patients and Methods

It was a prospective descriptive study from January, 1st 2016 to December 31, 2021. Children aged 0 to 15 years managed for testicular or para-testicular tumours were included. We recorded and analyzed medical data, and patients were followed up at a mean of 4.3 years.

After clinical examination, the dosage of tumour blood markers was systematic, as well as the inguinal-scrotum Ultrasound (US). In addition, thoracoabdominal and pelvis CT-scan were performed to look for metastases. Patients with testicular tumours underwent an inguinal orchiectomy and tumour resection for those with para-testicular tumours. Adjuvant chemotherapy or associated radiotherapy was introduced selectively following the histological finding and the international recommendations for testicular or para-testicular management of tumours in children. The follow-up was focused on clinical examination, the values of tumour blood markers, and imaging findings (CT-scan, MRI, bone scintigraphy). Data were recorded and analyzed by Epi info 7.6. We respect ethical principles.

Results

Thirteen patients met our criteria. Two tables summaries patient's characteristics. Table 1, patient's clinic and imaging data, and Table 2 the evolution.

The mean age was five years [16 days-14 years], and eight patients had an age ≤ 2 years (Table 1). No cancer history was recorded. Otherwise, one patient had contralateral cryptorchidism; another one underwent surgery for Fallot's teratology during the neonatal period, and one for renal lithiasis. Eleven patients were admitted on an emergency mode for acute bursal swelling, one for painless inguinal mass. It was a painless unilateral testicular mass, without any local inflammatory signs. The contralateral testicle appears normal. Symptoms evolve from a few days to several months [16 days to 14 months] (Table 1).

There were nine testicular tumours and four para-testicular tumours. The right side was affected in eight cases (Table 1), and none of our patients had signs of puberty. Ultrasonography found a heterogenous testicular mass, hyper vascularized sometimes with a mean diameter of 37.33 mm (Table 1).

There was no métastase at the time of diagnosis. The dosage of the ß-HCG and the α-FP was normal in six patients; the α-FP was higher than the physiologic values in six cases, and, in the last case, both the ß-HCG and the α-FP were higher (Table 2).

Eleven patients underwent uneventful primary orchiectomy by inguinal way. One patient had a tumour resection, a secondary orchiectomy for local recurrence, and the last one had only a tumour resection of twelve orchiectomies (Table 2).

After surgery, serum tumour markers became normal in four patients, in regression to the three others and remained normal in the six last patients (Table 2). Yolk sac tumour was the most frequent, followed by the mature teratoma (Table 2).

Only the patients with high-risk tumours received adjuvant chemotherapy. It was the RMS 2005 protocol for the

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Table 1: Characteristics of patient's testicular or para-testicular tumor in children.

	Age	Consult. Period	Side	US mensurations	Histologic type
Patient 1	16 days	16 days	Right	37 × 33 mm	Yolk sac tumor
Patient 2	2 years	20 days	Left	30 × 13 mm	Yolk sac tumor
Patient 3	9 months	2 days	Left	30 × 40 mm	Yolk sac tumor
Patient 4	14 years	10 years	Left	41 × 28 × 35 mm	Mature Teratoma
Patient 5	14 years	Discovery	Left	Normal	Mature Teratoma
Patient 6	14 years	20 days	Right	39 × 35 mm	Mature Teratoma
Patient 7	14 months	8 months	Right	Normal	Géant cell Fibroblastoma of the spermatic cord
Patient 8	2 years	14 months	Right	31 × 21 mm	Mature Teratoma
Patient 9	5 months	7 days	Right	30 × 23 mm	Yolk sac tumor
Patient 10	1 year	15 days	Left	Normal	High risk Yolk sac tumor
Patient 11	12 years	2 months	Right	27 × 17 mm	Alveolar Rhabdomyosarcoma high risk
Patient 12	9 months	1 months	Right	21 × 12 × 26 mm	Hamartoma fibrosis
Patient 13	5 years	2 months	Right	Normal	Embryonic Rhabdomyosarcoma high risk

Table 2: results of blood cell markers (AFP et B-HCG en UI/L) and their follow-up after surgery according patient' age and histologic founding.

	Age	Consult. Period	Before surgery	Histologic type	After surgery
Patient 1	16 days	16 days	AFP: 13221.2	Yolk sac tumor	AFP: 19.60
			B-HCG: 5650		B-HCG: 6.21
Patient 2	2 years	20 days	AFP: 18080	Yolk sac tumor	AFP: 4
			B-HCG: normal		B-HCG: normal
Patient 3	9 months	2 days	AFP: 1000	Yolk sac tumor	AFP: 4.79
			B-HCG: normal		B-HCG: normal
Patient 4	14 years	10 years	AFP: normal	Mature Teratoma	AFP: normal
			B-HCG: normal		B-HCG: normal
Patient 5	14 years	Discovery	AFP: normal	Mature Teratoma	AFP: normal
			B-HCG: normal		B-HCG: normal
Patient 6	14 years	20 days	AFP: normal	Mature Teratoma	AFP: normal
			B-HCG: normal		B-HCG: normal
Patient 7	14 months	8 months	AFP: normal	Géant cell Fibroblastoma of the spermatic cord	AFP: normal
			B-HCG: normal		B-HCG: normal
Patient 8	2 years	14 months	AFP: normal	Mature Teratoma	AFP: normal
			B-HCG: normal		B-HCG: normal
Patient 9	5 months	7 days	AFP: 400	Yolk sac tumor	AFP: 4.38 /14.2
			B-HCG: normal		B-HCG: normal
Patient 10	1 year	15 days	AFP: 28000	High risk Yolk sac tumor	AFP: 3.6/62.8
			B-HCG: normal		B-HCG: normal
Patient 11	12 years	2 months	AFP: normal	Alveolar Rhabdomyosarcoma high risk	AFP: normal
			B-HCG: normal		B-HCG: normal
Patient 12	9 months	1 months	AFP: 16990	Hamartoma fibrosis	AFP: 14.85
			B-HCG: normal		B-HCG: normal
Patient 13	5 years	2 months	AFP: normal	Embryonic Rhabdomyosarcoma high risk	AFP: normal
			B-HCG: normal		B-HCG: normal

rhabdomyosarcoma and TGM 95 protocol for the yolk sac tumour. One received eight cures, and the other one had 12 cures of IVA.

During the follow-up, three patients had complications: one retro peritoneum metastasis, one local recurrence and one local progression affection the penis and urethral. Second look surgery was then done for to patients. Fractional radiotherapy at 1.8 Gy/Fr was added for the two cases of rhabdomyosarcoma after gonadal transposition (Table 2).

After a mean period of follow-up of 4.3 years [1-6 years]:

- One patient presents two years after orchiectomy with acute lymphoid leukemia, treated and declared well from cancer after chemotherapy. He is still under follow-up and has no recurrence at five years.
- Twelve are under follow-up without any recurrence. Three chemotherapy protocols are finished, even the radiotherapy. One patient had the gonads repositioned, and this one is scheduled for the second one.
- The patient with local progression was lost on follow-up after one year.

Discussion

Testicular and para-testicular are rare in children. In Tunisia, only 12 cases were recorded over 27-years of experience. In our institution, 13 cases were recorded over six years [1,4]. However, their incidence is increasing [5]. Age at diagnosis varies, with a peak at the age of 2 years. Our patient's mean age was 5-yers that meets the literature data [6-8]. Just like Carlos OR, et al. [9], we found neonatal forms.

Symptoms are marked by painless testicular mass; in a few cases, acute testicular pain and exceptional incident discovery, as we and Sellami et al. [4,8] report. The right side is more involved, and Fadi et al. [8] report 23% of bilateral involvement.

The α-FP is high in over 90% of yolk sac tumours and 30.4% for immature teratomas. The ß-HCG is high at 2.2% for yolk sac tumours and 2.7% for immature teratomas [1]. Glenda Scandura, et al. [10] rapport that only 16 over 81 malignant tumours had an elevation of tumour blood markers. In our cases, the α-FP was high in 6 cases. Under the age of 14 years, yolk sac tumours are far the most frequent at 43.5% [4,11].

Among the para-testicular tumours, the rhabdomyosarcoma had

a particular aggressiveness. Therefore, early diagnosis and quality of management determine the prognostic [12]. This aggressiveness is confirmed in our two patients, as well as the predominance of the embryonic form before 10- years old and the alveolar form after 10 years old [12].

For the radio-histologic correlation, for a tumour less than 10 mm in diameter, there are 69% of benign tumours [10]. In our cases, both were more than 10 mm in diameter. Inguinal radical orchiectomy is the historic therapeutic approach for testicular tumours. However, 48% to 78% of benign testicular tumour reporters in children mad the conservative surgery increasingly promoted in the literature [1,13].

In the meta-analysis of Josias BG [7] over 88 publications, inguinal orchiectomy was performed in 87% and tumour resection with testicular conservation in 9%. Conservative surgery does not respect the rules of oncologic surgery. It is indicated for adequate remaining testicular healthy parenchyma and standard tumour blood cell markers [14]. Nevertheless, standard blood cell markers do never formally exclude a malignant tumour.

Inguinal orchiectomy remains the gold standard in our institution. Chemotherapy is selectively indicated following the histologic founding.

- For rhabdomyosarcoma, polychemotherapy is indicated for 18 to 24 months. These protocols are VAC, IVA and VIE (V: vincristine; A: actinomycin D; E: Etoposide; I: ifosfamide; C: cyclophosphamide) [12]. The IVA was the one done for our patients.
- The TG M95 protocol and the association VIP (Etoposide, ifosfamide and cisplatin) were used for the Yolk sac tumour.

The local recurrence and metastases are the risk for testicular and para-testicular tumours in children [7]. These complications were noticed in three patients. After conservative surgery, the rate of recurrence varies from 1.1 to 5%, and testicular atrophy from 0 et 0.37% [7,14,15]. No case of death was reported; one patient lost of follow-up. Five years of survival for non-metastatic tumours is 97.4% [11].

Conclusion

Testicular and para-testicular tumours are rare in children, and germ cell tumours are the most frequent. The diagnosis approach is well codified. Inguinal orchiectomy is the gold standard of management. Although the prognostic is good, patients should be followed-up till adulthood.

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