Scientific Research Publishing

Open Journal of Pediatrics, 2023, 13, 207-213
https://www.scirp.org/journal/ojped
ISSN Online: 2160-8776
ISSN Print: 2160-8741

The Pre-Diagnosis History of Gynecologic Tumors in Children

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How to cite this paper: Alaoui, O., Baldé, F.B., Mahmoudi, A., Khattala, K. and Youssef, B. (2023) The Pre-Diagnosis History of Gynecologic Tumors in Children. *Open Journal of Pediatrics*, **13**, 207-213.
https://doi.org/10.4236/ojped.2023.132026

Received: December 28, 2022
Accepted: March 11, 2023
Published: March 14, 2023

Abstract

Introduction: Gynecologic tumors represent less than 5% of all solid tumors in children. **Patients and Method:** Through a prospective and descriptive study over 4-years, we included girls aged less than 15 years admitted for a gynecologic tumor. We collected and analyzed the medical data, and paid particular attention to the pre-diagnosis history. **Results:** Eleven girls met our criteria with a mean age of 8.5 years (2 - 13 years). Abdominal pain, abdominal distension or protrusion of a vaginal mass was the main symptoms. The patients were seen at least by one doctor outside the surgical structure [1] [2] [3]. Four of eleven were received by two different doctors before carrying out the evocative radiological assessment. The average consultation delay was 7-months (24 hours to 18 months). There were nine ovarian tumors and two vaginal tumors. The histological study revealed five of eleven (45%) malignant tumors. **Conclusion:** Gynecological tumors in children are rare. However, there is a significant proportion of malignant tumors. All doctors can be confronted with it and should not delay the radiological assessment.

Keywords

Gynecologic Tumor, Diagnostic, Children

1. Introduction

Gynecologic tumors represent less than 5% of all solid tumors in children. They are predominated by ovarian tumors which are benign in majority [1]. However, it is always necessary to keep on mind the malignancy possibility and undertake diagnosis approach on this direction. Surgery, surgery-chemotherapy or surgery-chemotherapy-surgery constitutes the keys of the management. In this study,

we aimed to report the pre-diagnosis history of gynecologic tumors in children.

2. Patients and Methods

It was a descriptive and prospective study over four years (January 1st, 2018 to December 31st, 2021). All the girls aged from 0 to 15 years admitted for gynecologic tumor were included. We secondary excluded the ovarian necrosis under ovarian torsion without any pathologic tissue on the histologic study. Were collected personal medical record and a we paid a particular attention on the pre diagnosis history. The outcome was followed-up at mean for 2.9 years. We performed a descriptive analysis of the patients. Qualitative variables were presented in pourcentage, and those quantitatives in mean and mode.

Ethic aspects: We respect Helsinki's principles to which adheres our institution.

3. Results

Eleven girls met our criteria. The mean age was 8, 5 years [2]-[13]. Modal ages were 5, 11 and 13 years. The reported symptoms were abdominal distension, abdominal pain, nausea, vomiting, diarrhea, protrusion of vaginal mass. About the pathologic history, two girls had started tir menstruations (*patients* 2 *and* 3), one of whom (*patient* 3) reported metrorrhagia. One other girl has been operated for spina bifida and hydrocephalus in the neonate period (*patient* 5).

The pre diagnosis history reveals that patients were seen at least by one doctor (pediatrician, generalist) outside the surgical structure [1] [2] [3]. Four to eleven were received by two different doctors before carrying out the evocative radiological assessment.

Almost half of the patients (five out of eleven) were referred from anther cities. The same proportion of patient was seen by private clinic before to be referred in our institution.

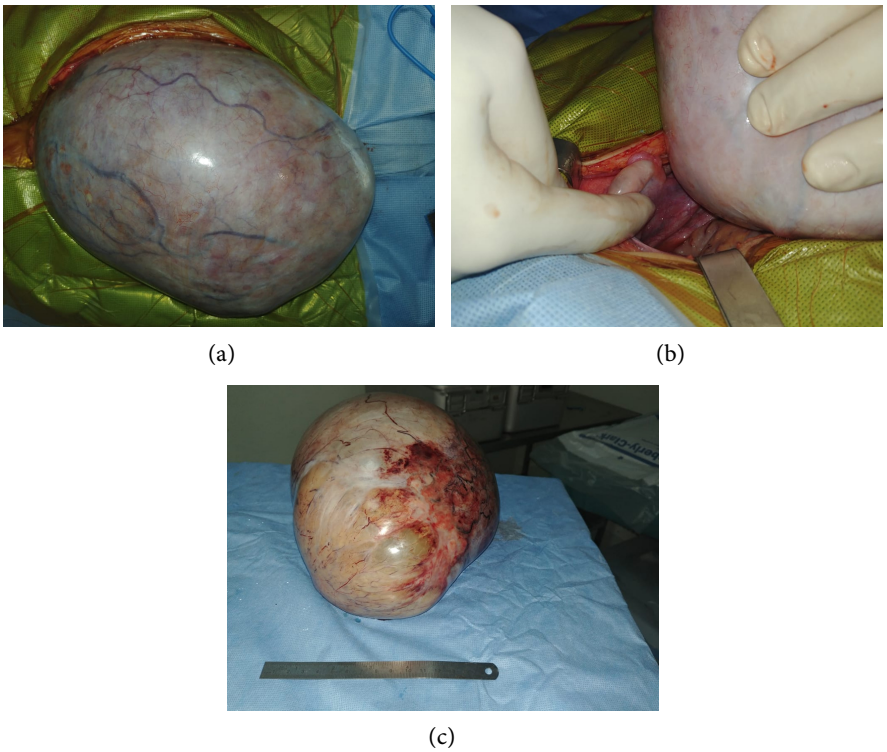
The average consultation time of 7-months (24 hours to 18 months). Four out to eleven girls were admitted with a typical picture of ovarian torsion (**Table 1**).

The clinical finding revealed the perception of an abdominal or abdominopelvic mass (**Figure 1(a)**, **Figure 1(b)** and **Figure 1(c)**), a vulvar mass (**Figure 2**), and abdominal tenderness. One patient also presented light brown birthmarks (*Patient* 6). Blood levels of alpha-fetoprotein (AFP) and beta choriogonadotropin hormone (β-HCG) performed in nine patients were abnormal in eight patients. One patient (*patient* 11) had elevated B-HCG at 25,428 ng/ml (VN < 0.1).

The abdominopelvic ultrasound performed on all the patients revealed an abdominal and or pelvic mass in nine patients (**Figure 3**) with an average diameter of 92*77 mm [40*31 - 183*136]. This one is supplemented by a CT scan and or abdominopelvic MRI. One patient had an invasion of the rectum with loss of the fatty border of separation (*patient* 6) and another lung metastases at the time of diagnosis (*patient* 5). We encountered nine ovarian masses (including six on the right) and two vaginal tumors.

Table 1. Clinical and therapeutic characteristics of the patients in our series.

	Age (year)	Consultation period	Histology	Surgery	Outcome
Patient 1	11	1 Years	Right ovarian mucinous adenocarcinoma	Oophorectomy	LFU
Patient 2	13	1 Years	Right ovarian dysgerminoma	Oophorectomy	LFU
Patient 3	13	1 Years	Left ovarian follicular cyst	Cystectomy	Normal
Patient 4	5	3 Months	Right dermoid ovarian cyst	Oophorectomy	Normal
Patient 5	3	5 days	Right immature ovarian teratoma	Oophorectomy	Chemotherapy
Patient 6	11	1 Years	Vaginal plexiform Neurofibroma	Tumor resection	LFU
Patient 7	5	1 Years	Left dermoid ovarian cyst	Oophorectomy	Normal
Patient 8	2	4 days	Right dermoid ovarian cyst	Oophorectomy	Normal
Patient 9	4	1 Years and 6 Months	vaginal Rhabdomyosarcoma	Palliative Surgery	Chemotherapy
Patient 10	11	2 Days	Right dermoid ovarian cyst	Oophorectomy	Normal
Patient 11	8	2 Months	Right ovarian Choriocarcinoma	Oophorectomy	Chemotherapy



(a) (b)

(c)

Figure 1. Per operative images of patient 1 (according to the Table 1) presenting an abdominal a pelvis masse. (a): voluminous right ovarian. (b): right ovarian masse a left ovarian has a normal macroscopic aspect. (c): the ovarian tumor after resection.

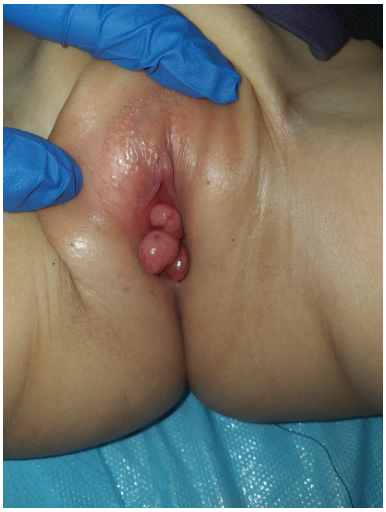


Figure 2. Clinical image of patient 9 (according to **Table 1**). Vaginal masse evolving since 18 months. Histologic study concluded to a vaginal rhabdomyosarcoma.

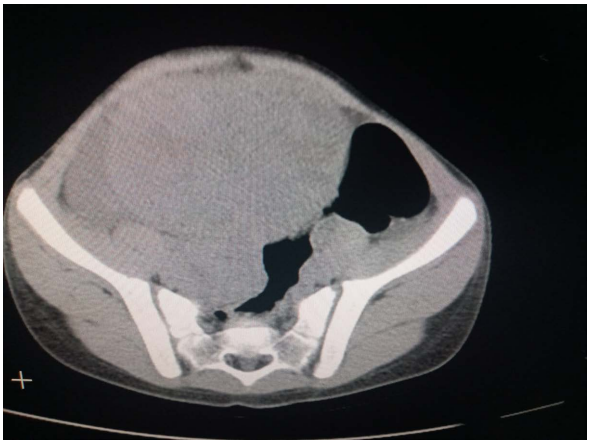


Figure 3. Abdominal CT-scanTDM of patiente 11 (according to **Table 1**). Right ovarian masse 141*87 mm de diameter. Histologic study concluded to a choriocarcinoma.

The time to surgery varied from a few hours to five days with the laparoscopic approach in 3 patients. Patients underwent uneventful surgery.

The histological study revealed five out of eleven (45%) malignant tumors (**Table 1**). They were ovarian dysgerminoma, ovarian mucinous adenocarcinoma, ovarian choriocarcinoma, immature ovarian teratoma, and vaginal rhabdomyo-sarcoma. Particularities were accounted for the patients 8 and 11. For patient 8 of our series, a fibro-epithelial polyp was evoked before being confirmed being a vaginal rhabdomyosarcoma. For patient 11, a border-line mucinous cystadenoma evocated before being confirming the ovarian choriocarcinoma.

The chemotherapy based on the TGM 95 protocol was initiated for two immature teratomas and choriocarcinoma. The MMT 2005 protocol for rhabdomyosarcoma.

As for the medium-term evolution, β-HCG became normal (25,428/9500/<2) after 16 courses of chemotherapy. One patient (Patient 9) presented a local recurrence for which palliative surgery was performed, and she is continuing her

chemotherapy (VIP 20). Finally, three out of eleven were lost to follow-up (LFU) after one year of monitoring, which was unremarkable (patients 1, 2 and 6); 9/11 are followed regularly in multidisciplinary (pediatric surgery, pediatric oncology, radiology).

4. Discussion

Gynecologic tumors in children are rare [2]. The mean age at diagnosis varies from one study to another: 8.5 years in our series 10.3 years in that of E. Péroux *et al.* [2].

The symptoms reported in our study corroborate those reported in the literature, and ovarian torsion was encountered in four out of eleven patients [2]. The risk of ovarian torsion on teratoma is between 3 to 16% [3].

Unilateral post-pubertal oophorectomy is known to be associated with an increase in FSH at the age of thirty-five (OR: 2.8; CI: 0.7 - 11.2) and with the occurrence of ovarian failure before forty years of age (OR: 4.3; CI: 0.9 - 20.4); but it is not known to what extent unilateral oophorectomy performed before puberty affects ovarian reserve. Of the eight oophorectomies in our study, only two patients were pubertal [4].

The consultation time in our series is close to some data in the literature. A maximum delay of at least 12 months for all is reported (**Table 2**). Ndungo K. *et al.* [5] report a delay of one year for a vulvar tumor. The same delay was observed in our patients who presented a vaginal tumor.

As for the pre-diagnostic history, Sarah Tatencloux *et al.* [8] report a median number of consultations of 2 [0 - 7] for children and adolescents with solid tumors. This number was smaller in our study 1 [1 - 3].

In the reported series, 10% of ovarian masses in children are malignant tumorous. A tumor larger than 8 cm has an OR (odds ratio) of malignancy of 19 (CI: 4.4 - 81.6) [9]. In our study, the mean diameter of ovarian tumors was more than 8 cm (92*77 mm). It could explain the high proportion of malignant tumors in our series (4/9 ovarian tumors).

Germ cell tumors are the most frequent of ovarian tumors in children and adolescents, where mature teratomas or benign dermoid cysts represent 55% - 70% of cases [10]. The proportion of germ cell tumors was seven out of nine, and four were dermoid cysts. E. Péroux *et al.* [2] found 14/42 (33%) malignant ovarian tumors. This proportion was 4/9 (44%) in our series.

Table 2. Comparison of the consultation time in our study with that in the literature.

	Place of study	Mean delay	Extremes
Our study	**Fès**	7 months	0 - 18 months
Hanane ZEROUAL [6]	**Rabat**	2 months	0 - 13 months
Fadoua FETTAL [7]	**Marrakech**	6 months	0 - 12 months

Among the malignant ovarian tumors, 45% were dysgerminomas [11], one case in our study. Other ovarian malignancies encountered in our series were mucinous adenocarcinoma, choriocarcinoma and immature teratoma.

Vaginal rhabdomyosarcoma is rare in the pediatric population. Over 20 years, 240 cases were reported in Europe [11] [12].

Surgery is becoming less and less invasive. Cystectomy is considered for a single cyst with no signs of malignancy (imaging and biology assessment) [13]. For ovarian tumors, conservative surgery by unilateral oophorectomy is the recommended option. However, some situations have been reported in the literature, such as the 10% bilateral involvement reported by Fadwa El Omrani [3]. There was no bilateral involvement in our study. Chemotherapy is well codified and is introduced according to the histological type.

For ovarian tumors, the recurrence rate is 27% after conservative surgery within 19 months [14]. No recurrence of ovarian tumors has been reported in our series so far. As for vaginal or vulvo-vaginal tumors, as in the literature, we have been confronted with this situation [5] [14].

The management of gynecological tumors in children remains an even greater challenge in developing countries. This multidisciplinary management is often regional, as in the case of Ndungo K. *et al.* [5] in Gabon, where a 3-year-old girl operated on for vulvar rhabdomyosarcoma was transferred to Uganda for further management due to the lack of local centres specialized in the management of children with cancer.

5. Conclusion

Gynecological tumors in children are rare. The delay of consultation remains long in the lesser symptomatic forms. Children consult at least one doctor before the diagnosis. Surgery remains a key means of management and should be as conservative as possible to preserve subsequent fertility.

Acknowledgements

We thank the teams of the pediatric oncology unit of the Hassan II University Hospital of Fez and the radiology department of the Hassan II University Hospital of Fez.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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