ORIGINAL ARTICLE

Clinico Pathological Review of Adnexal Masses in Children and Adolescents at a University Hospital

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Abstract

Background & Objective: The study objective was to review the clinic-pathological characteristics of adnexal masses in the children and adolescent population in our institution.

Methods: Retrospective analysis of 178 adolescent patients including neonates, presenting with adnexal masses between January 2000-December 2012, was performed.

Results: Records of a total of 178 patients were analyzed. The mean age was 14 years. The most common symptom was abdominal pain noted in 146(82%) patients followed by nausea and vomiting in 28(15.7%) and incidental findings in 19(10.7%). The majority of the ovarian masses were benign. The follicular cyst was the most common histopathological type in 66 patients (37.1%) followed by a hemorrhagic cyst in 40(22.5%), benign teratoma in 21(11.8%), benign serous cyst in 20 patients (11.2%), endometrioma and mucinous cystadenoma in 13 patients (7.3%). The average tumor size observed in the present study was 7.84cms. However, the size varied in different age groups such as 4.27 cm size in neonates, and 7.1 cm in 1-14 years age group, and 8.61 cm in 15-19 years age group with a P-value of 0.009. Out of a total of 178 patients, 46(25.8%) were managed conservatively. However, 90(50.6%) patients underwent ovarian cystectomy, 26(14.6%) were managed by unilateral salpingo-oophorectomy, 10(5.6%) by unilateral oophorectomy. Aspiration and biopsy were done in 6(3.4%) patients.

Conclusion: Incidence of adnexal masses increases with age with maximum cases between age 14 and above. Adolescent girls with adnexal masses can have variable presentations however abdominal pain is the most common symptom. Mostly these are benign masses therefore conservative approach for treatment should be adopted.

Keywords: *Adnexal diseases, adolescent, benign, malignant, children.*

INTRODUCTION

Diagnosing and treating adnexal masses in children and adolescents is a big challenge owing to fertility concerns in young females, lack of awareness and limited health facilities. Gynecologic tumors are infrequent in children, constituting 5% of all the pediatrics neoplasms [1]. The actual incidence of pediatric ovarian masses is unknown; About one-half of all adnexal masses in children are ovarian in origin and estimated to occur at a rate of approximately 2.6 cases per 100,000 girls per year, while malignant ovarian neoplasms make about 1% of all childhood cancers [2].

In the adolescent age group, malignant ovarian masses are rare, lethal and notorious and a lot of controversies are there regarding their management [3]. Commonly patients with an adnexal mass present with sudden intense pelvic or lower abdominal pain secondary to torsion of the adnexa or hemorrhage into the lesion [4]. Sometimes fever, nausea and vomiting can also occur [5]. Other presentations can be mild, chronic pain with gradually increasing abdominal girth, precocious puberty due to hormone-producing tumors and, menstrual irregularities [6].

Various classification and nomenclature systems for ovarian masses exist. These can be neoplastic and non-neoplastic. Non-neoplastic conditions include follicular cysts, corpus luteal cyst, endometriotic cyst and hemorragic cyst. Intraoperative and postoperative management varies according to the age of the patient, size of the lesion and whether it is primarily cystic or solid cysts [7-9], and endometriomas. Neoplastic processes include both benign tumors such as mature cystic teratomas as well as highly malignant tumors. Three main categories for these tumors are: epithelial, stromal, and germ cell tumors. The predominance of germ cell tumors is noted as per previous data [7]. In addition, there are tumors of low malignant potential that frequently follow a benign clinical course [8] if timely and optimal surgery is done.

Therefore, more information about the clinical characteristics of adnexal masses in adolescent females is required to establish procedures for early diagnoses and management of such lesions in this age group. The aim of this study is to review the clinicopathological characteristics of adnexal masses in the children and adolescent populations in our institution.

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Received: November 06, 2020; Revised: July 13, 2021; Accepted: July 26, 2021
DOI: https://doi.org/10.37184/lnjcc.2789-0112.2.4

MATERIALS AND METHODS

This is a retrospective observational study conducted at Aga Khan University Hospital. We reviewed the previous records of 12 years from January 2000–December 2012. All patients with age less than 20 years including neonates who presented with adnexal masses at Aga Khan University Hospital Karachi, Pakistan from January 2000 –December 2012 were included in the study. Patients with recurrent ovarian masses were also included. However, females with germ cell tumors at sites other than ovary, male patients with germ cell tumors and adnexal masses in females of more than 20 years of age were excluded. Other variables that were recorded for evaluation included: Age, clinical presentation, radiological findings (U/S, MRI/CT scan pelvis), histopathological findings, primary treatment method-conservative or surgery.

The statistical software of SPSS (version 19) was used to analyze the data. Mean and standard deviations for continuous variables and proportions for categorical variables were estimated. The statistical analysis was done by using the student's t-test/Anova and chi-square test.

RESULTS

The present study included 178 patients with adnexal mass varying in age from the neonatal period till 20 years. The mean age of the patients was 14.48+5.17 years. Out of the total sample, 16(9%) were neonates with adnexal masses, 47(26.4%) patients were between one year to fourteen years of age, and 115(64.6%) patients were between fifteen to nineteen years of age. Abdominal pain was the most common presenting symptom noted in 146(82%) patients followed by nausea and vomiting in 28(15.7%) patients and as incidental finding in 19(10.7%) patients. Other presenting complaints reported were; abdominal distension 16(9%), menstrual irregularities 12(6.7%), constipation 3(1.7%), urinary retention 2(1.1%) and loss of appetite 1(0.6%).

The majority of the ovarian masses were benign. The follicular cyst was the most common benign pathology found in 66 patients (37.1%) followed by hemorrhagic cyst found in 40 patients (22.5%), benign teratoma found in 21 patients (11.8%), benign serous cyst found in 20 patients (11.2%), endometrioma and mucinous cystadenoma found in 13 patients (7.3%). Moreover, 1 patient (0.6%) had fibroma, benign sclerosing tumour, immature teratoma, dysgerminoma and borderline tumor. However, out of 16(9%) neonates, 11(68.8%) had a follicular cyst and 5(31.3%) had a hemorrhagic cyst. The same was the observation in the 1-14years age group in which follicular cyst was noted in 20(42.6%) patients out of 47 patients of this age group and the hemorrhagic cyst was observed in 13(27.7%) patients. Similarly, in the age group 15-19 years follicular cyst was noted in 35(30.4%) patients and hemorrhagic cyst in 22(19.1%) out of a total of 115 patients in this age group (Table **1**).

Table 1: Clinico pathological characteristics of adnexal masses in children and Adolescent population (n=178).

Variables	Point Estimation	Percentage or [Min-Max]
Age (Years)	14.48±5.17	[0 to 19]
Presenting Symptom		
Abdominal pain	146	82
Nausea / Vomiting	28	15.7
Loss of Appetite	1	0.6
Urinary retention	2	1.1
Abdominal distension	16	9
Weight loss	0	0
Constipation	3	1.7
Menstrual irregularity	12	6.7
Incidental finding	19	10.7
Histological Type		
Simple /follicular cyst	66	37.1
Benign teratoma	21	11.8
Immature Teratoma	1	0.6
Dysgerminoma	1	0.6
Endometrioma	13	7.3
Benign serous cyst	20	11.2
Benign mucinous cyst adenoma	13	7.3
Boderline tumors	1	0.6
Haemorrhagic cyst	40	22.5
Benign sclerosing tumor	1	0.6
Fibroma	1	0.6
Tumor Size (cm)	7.84±5.58	[0 to 33.3]
Appearance		
Cystic	151	84.8
Solid	3	1.7
Solid and Cystic	24	13.5
Torsion	3	1.7
Laterality		
Unilateral	155	87.1
Bilateral	23	12.9
Ascites	45	25.3
Minimal	30	16.9
Mild	12	6.7
Moderate	4	2.2
Treatment		
Conservative management	48	27
Laparoscopic surgery	52	29
Laparotomy	78	44

The average size of the tumor observed in the present study was 7.84+ 5.58cm. However, variation in size was noted in different age groups such as 4.27+1.67cm size in neonates, and 7.16+5.24cm in the 1-14years age group, and 8.61+5.86cm in the 15-19years age group with a P-value of 0.009. Out of 178 patients 151(84.8%) presented with cystic mass, while 3(1.7%) had solid mass, and 24(13.5%) had solid cum cystic mass. Torsion was noted in 3(1.7%) patients; 2(4.3%) in the 1-14years age group, and 1(0.9%) in the 15-19years age group. Unilateral cysts were noted in 155(87.1%) patients whereas 23(12.9%) patients had a bilateral cyst. Finally, out of 178 patients, ascites were noted in 45(25.3%) patients.

About 27%(46) of these patients did not require any surgical intervention and were managed conservatively. About 29% of patients underwent laparoscopic surgery as compared to 44% who underwent laparotomy. The rate of laparoscopic surgery was higher among gynaecologists (38%) versus paediatric surgeons (9%). 90(50%) patients underwent ovarian cystectomy,

10(5.6%) had unilateral oophorectomy and 26(14.5%) underwent unilateral salpingo-oophorectomy (Table **2**).

Table 2: Comparison of clinic opathological characteristics of adnexal masses in children and Adolescent population (n=178).

Variables	Age groups			P-Values
	Neonate [n=16]	1 to 14 Years [n=47]	15 to 19 Years [n=115]	
Presenting Symptom				
Abdominal pain	2(12.5%)	43(91.5%)	101(87.8%)	0.0005
Nausea / Vomiting	1(6.3%)	7(14.9%)	20(17.4%)	0.509
Loss of Appetite	0(0%)	0(0%)	1(0.9%)	0.759
Urinary retention	0(0%)	0(0%)	2(1.7%)	0.575
Abdominal distension	2(12.5%)	4(8.5%)	10(8.7%)	0.875
Constipation	0(0%)	1(2.1%)	2(1.7%)	0.847
Menstrual irregularity	-	3(6.4%)	9(7.8%)	0.501
Incidental finding	13(81.3%)	1(2.1%)	5(4.3%)	0.0005
Histological type				
Simple /follicular cyst				
Benign teratoma	11(68.8%)	20(42.6%)	35(30.4%)	0.008
Immature Teratoma	0(0%)	5(10.6%)	16(13.9%)	0.260
Dysgerminoma	0(0%)	1(2.1%)	0(0%)	0.246
Endometrioma	0(0%)	0(0%)	1(0.9%)	0.759
Benign serous cyst	0(0%)	0(0%)	13(11.3%)	0.021
Benign mucinous cystadenoma	0(0%)	5(10.6%)	15(13%)	0.298
Borderline tumors	0(0%)	3(6.4%)	10(8.7%)	0.439
Hemorrhagic cyst	0(0%)	0(0%)	1(0.9%)	0.759
Benign sclerosing tumor	5(31.3%)	13(27.7%)	22(19.1%)	0.338
Fibroma	0(0%)	0(0%)	1(0.9%)	0.759
	0(0%)	0(0%)	1(0.9%)	0.759
Tumor Size (cm)	4.27±1.67	7.16±5.24	8.61±5.86	0.009‡
Appearance				0.685
Cystic	14(87.5%)	42(89.4%)	95(82.6%)	
Solid	0(0%)	0(0%)	3(2.6%)	
Solid and Cystic	2(12.5%)	5(10.6%)	17(14.8%)	
Torsion	0(0%)	2(4.3%)	1(0.9%)	0.271
Laterality				0.105
Unilateral	16(100%)	43(91.5%)	96(83.5%)	
Bilateral	0(0%)	4(8.5%)	19(16.5%)	
Ascites	0(0%)	14(29.8%)	32(27.8%)	0.045
Minimal		12	18	
Mild		2	10	
Moderate		0	4	

DISCUSSION

Ovarian masses are a very rare occurrence in children, with an approximated prevalence of 2.6 cases per 100,000 girls of neoplastic ovarian masses each year [10]. Tumors occurring during childhood and premenarchal age are non-neoplastic [11].

In this study, we have analyzed the diagnosis and treatment of 178 patients with an adnexal mass. The mean age of the patients included in the study was 14.48+5.17 which is consistent with existing data as Herman *et al.* reported the mean age to be 10.2+5.6 years at the time of first consultation [10].

Symptom manifestation of adnexal masses can vary. Abdominal pain is the most common symptom in young patients [11]. However, abdominal pain is considered an atypical symptom in 30-50% of young females and is difficult to differentiate from acute appendicitis. Therefore, ultrasound should be performed in females presenting with the symptoms of abdominal pain to rule out other possibilities [12]. According to Rossi BV *et al.* acute onset of unilateral abdominal pain, mostly on the right side with nausea and vomiting is the commonest presentation [13]. Other presenting symptoms are an incidental finding on a routine scan, abdominal distension, menstrual irregularity, urinary retention, constipation and loss of appetite.

In the present study, 176 patients out of a total of 178 had benign histopathology, while one patient had a borderline tumor, and 1 had immature teratoma. Herman *et al.* had also reported in their study that the majority of adnexal masses in children are benign [14].

Ovarian torsion is commonly associated with dermoid cyst and our study supports this observation as 12% of patients had dermoid cyst and torsion of the cyst was noted in 4.7% of cases. In pediatric patients, a longer infundibulopelvic ligament leads to ovarian torsion more frequently [15]. Somehow ovarian torsion is more on

the right side and is commonly misdiagnosed as acute appendicitis [16, 17]. It is still not clear whether left lesions are commonly missed or sigmoid colon helps prevent left adnexal torsion [2, 18].

Ovarian tumors are usually benign in females less than 5 years old. Cystic ovarian tumors are commonly seen in the pediatric population. Ovarian tumors must be considered one of the differential diagnoses of young girls with abdominal pain, mass or other non-specific symptoms [11]. Templeman *et al.* stated that malignant ovarian tumors in children and adolescents are rare, accounting for 0.9% of all malignancies in this age group [19]. Hassan *et al.* shared similar findings narrating that during the first two decades of life the most frequent tumors found in the female genital tract are benign [20]. Both these studies are consistent with our study as we had only one immature teratoma (0.6%) and one borderline tumor (0.6%) out of 178 patients. The rest of the patients had benign histopathologies.

The most common histopathological finding in our study was a simple follicular cyst, second, it was a hemorrhagic cyst followed by benign teratoma (dermoid cyst), benign serous cystadenoma, endometrioma and benign mucinous cystadenoma respectively. Cassandra M. Kelleher asserted that simple cysts are the most common adnexal masses in pediatric age groups and represent functional cysts in the vast majority of patients [6].

In neonates, only simple follicular cyst and hemorrhagic ovarian cyst were the histopathological findings either found incidentally on antenatal ultrasound or diagnosed later on. This is consistent with the findings of Khedkar *et al.* that follicular cysts are common in fetuses and neonates and their frequency increases with advancing gestational age. These are commonly detected on antenatal ultrasound examinations [11]. There is uncertainty about Etiology, but the most probable explanation is that it arises from ovarian stimulation by maternal and fetal gonadotropins [21].

Qublan HS also recommended conservative therapy with serial ultrasounds for simple asymptomatic 2 to 5 cm cyst because of the likely possibility of spontaneous regression within 4 to 5 weeks [22]. Functional cysts greater than 5cm can take up to 03 months to resolve completely [23].

CONCLUSION

Adnexal masses in younger age groups have variable clinical presentations and mostly benign pathologies. Conservative management, minimal access surgeries and fertility preservation surgeries are the best management options in this group of patients.

ETHICS APPROVAL

This article does not contain any studies performed on human subjects as it was a retrospective study done after institutional ethical board approval.

CONSENT FOR PUBLICATION

Not applicable

FUNDING

None.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

ACKNOWLEDGEMENTS

None.

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QUIZ AND KEYS

Hira Khan Afridi

QUESTION # 1

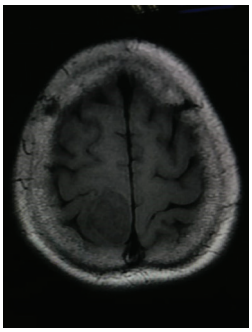

Fig. (a)

Fig. (b)

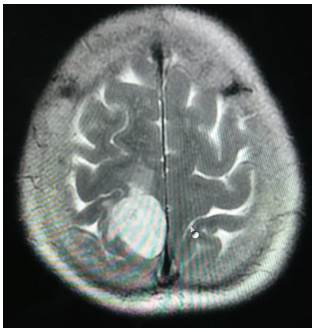

Fig. (c)

a. What is the diagnosis?

b. This patient had this finding, size of lesion being 2-2.5 cm. How will you manage?

QUESTION # 2

70 year old male patient, diagnosed as chronic lymphocytic leukemia with del 17p found. His Absolute lymphocyte count (ALC) was 20,000 when he was started treatment with Bruton Tyrosine Kinase Inhibitor (IBRUTINIB). Patient after 1 month of treatment came with CBC showing ALC of 25,000.

How shall this patient be managed further?

(See page 63 for answer keys)