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Shift towards minimally invasive surgery of adnexal pathologies in children: A predictive model study

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Abstract. – OBJECTIVE: The purpose of this article is to investigate these difficulties and to provide a modernized and comprehensive understanding of the diagnosis, management, and long-term outcomes of adnexal masses in pediatrics.

SUBJECTS AND METHODS: This study retrospectively reviewed the medical records of 428 patients who were diagnosed with adnexal pathologies and underwent Surgery.

RESULTS: In pre-adolescents, the duration of symptoms shows a statistically significant positive correlation with lesion size (p=0.006, r=0.621). The duration of symptoms shows a statistically significant positive correlation with lesion size (p<0.001, r=0.460). The formula is a mathematical model developed to predict the size of a lesion (a cyst or mass) in centimeters, based on the duration of the patient's symptoms in months. For mass size, the optimal cut-off was found to be 4.5 cm. This value yielded a higher AUC of 0.85 (95% CI:0.74-0.96), with a sensitivity of 94% and a specificity of 55%.

CONCLUSIONS: This study on adnexal pathologies in children indicates a shift towards minimally invasive laparoscopic surgery. A novel model predicting lesion size based on symptom duration was developed, and surgical intervention thresholds were established.

Key Words:
Adnexal pathologies, Laparoscopy, Symptoms, Prediction model, Mass size.

Introduction

Adnexal masses, which account for only 1% to 2% of all masses found in children, represent a complex medical scenario that requires intervention that is both prompt and careful[1]. The fact that these masses, which originate in the ovary, are benign in approximately sixty to seventy percent of cases highlights a significant challenge in the field of healthcare. The 2.7% of all gynecological emergencies are caused by them, particularly due to the risk of torsion, which requires a delicate balance between a prompt response and a precise approach[2,3]. In today's technologically advanced medical landscape, imaging tools can diagnose and treat tumors, cysts, and torsions in individuals with symptoms[4].

In cases of adnexal torsions, the importance of early diagnosis and intervention cannot be overstated. The incidence rate for this condition is reported to be between 2 and 4.9 per 100,000 people[5]. When it comes to preventing the loss of ovarian reserve, timely intervention is of the utmost importance. Due to the fact that these diseases manifest themselves in childhood, the treatment strategy is of the utmost importance in order to protect the child's future physical development and fertility. In order to effectively manage these conditions, it is necessary to protect the ovarian tissue whenever this is possible and to keep the hormonal activity stable[6].

The purpose of this article is to investigate these difficulties and to provide a modernized and comprehensive understanding of the diagnosis, management, and long-term outcomes of adnexal masses in pediatrics.

Subjects and Methods

Patients and Study Design

This study retrospectively reviewed the medical records of 428 patients who were diagnosed with adnexal pathologies and underwent surgery at Başakşehir Çam and Sakura Training Hospital between January 2015 and December 2022. After excluding incomplete or missing data, a total of 428 patients were included in the study. The patient's age, whether the lesion was a cyst or tu-

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mor, lesion side, surgical technique, incision type, treatment method, and whether enucleation was performed were extracted from the retrospective data. A new model was developed to evaluate the relationship between patients' symptoms and cyst size and to predict cyst size based on the patient's symptoms. The correlation between the cutoff value for performing laparotomy and its correlation with symptom duration was investigated.

Inclusion Criteria

Patients with complete retrospective data and accessible records were included in the study.

Exclusion Criteria

Patients with incomplete or unavailable retrospective data were excluded from the study. Additionally, cases that involved intrauterine-diagnosed ovarian pathologies, ruptured hemorrhagic cysts, and cases with positive findings suggestive of torsion on Doppler ultrasound but negative findings on surgical exploration, were not included. Furthermore, patients who could not obtain informed consent for surgery and those who wanted to continue their follow-up at different centers due to social reasons were also excluded from the study.

Ethical Approval

The ethics committee approval for this study was obtained from the Non-Interventional Local Ethics Committee of Başakşehir Çam and Sakura City Hospital (number: KAEK/2023.05.191). Informed consent was obtained from all patients.

Limitations of the Study

The retrospective nature of the study may limit the accuracy and comprehensiveness of the collected data due to incomplete medical records or possible recall bias. As the study is conducted at two institutions in Turkey, the findings may not be generalized to other settings or populations. Regional variation in healthcare practices and population characteristics could influence the outcomes. The study did not report any follow-up data. Therefore, long-term outcomes or complications could not be assessed. The pathologies result was not evaluated, which limits the understanding of the diagnostic accuracy and the precise nature of the adnexal pathologies.

Strength of the Study

We developed a mathematical model to predict lesion size based on the duration of symptoms, which could potentially assist in the management of these patients. The study determined cut-off values for cyst, mass, and lesion sizes, which can be used to guide the choice between laparoscopy and laparotomy. This is a practical contribution that may aid decision-making in clinical practice.

Statistical Analysis

The patient data was subject to a thorough statistical analysis, which included evaluating descriptive statistics (mean standard deviation [SD] or median [IQR]), frequencies, and other attributes across all categories. The continuous data were presented as mean plus or minus standard deviation. The Shapiro-Wilk and Kolmogorov-Smirnov tests were used to determine the normality of the distribution of continuous variables. For comparing continuous variables, the Student's *t*-test was used for normally distributed data, while non-parametric tests were used for data that did not have a normal distribution. For comparing categorical variables, the Chi-square test was used. To evaluate cut-off analysis, the Receiver Operating Characteristic (ROC) analysis was used. The Spearmen correlation test was used to determine the relationship between chemerin levels and tumor size. SPSS Statistics for Windows, Version 24.0 (IBM Corp., Armonk, NY, USA) was used for all statistical analyses. The *p*-values were two-sided, with 0.05 being considered statistically significant.

Results

In this study, we evaluated the characteristics and surgical approaches for adnexal pathologies in a cohort of 428 pediatric patients. Pre-adolescents constituted 30.8% (n=132). The causes of adnexal pathologies were primarily due to cysts, accounting for 67.1% (n=287) of the cases. Spontaneous adnexal torsion was the second most common cause, representing 18.7% (n=80) of the cases. Masses were responsible for 14.3% (n=61) of the cases. When examining the location of the lesion, most were found on the right side, making up 53.0% (n=227) of the cases. The left side had 44.6% (n=191) of the cases. A smaller number, 2.3% (n=10), had lesions on both sides. Of all the surgeries performed, the preferred surgical approach was laparoscopy, used in 66.8% (n=286) of the cases. The other 33.2% (n=142) underwent a different procedure called laparotomy. The 3-port method was the most common technique employed in 67.1% (n=287) of the surgeries. A large majority of the surgeries, 84.3% (n=361), were

Table I. General Characteristics of the patients.

Characteristics	N	%
Age Period		
Pre-Adolescent	132	30.80
Adolescent	296	69.20
Etiology		
Cyst	287	67.1
Mass	61	14.30
Spontaneous adnexal torsion	80	18.70
Side		
Right	227	53.0
Left	191	44.6
Bilateral	10	2.30
Surgical approach		
Laparoscopy	286	66.8
Laparotomy	142	33.2
Incision		
3 Port	287	67.1
Midline	40	9.30
Pfannenstiel	101	23.60
Procedures		
Salpingo-oophorectomy	67	15.70
Ovarian sparing surgery	361	84.30
Cyst/mass enucleation		
Yes	278	65.0
No	150	35.0

ovarian-sparing. In 15.7% (n=67) of the cases, salpingo-oophorectomies were performed. Additionally, enucleation was conducted in 65.0% (n=278) of the patients (Table I).

Of the patients in our study, 65% (n=278) primarily reported abdominal pain as their initial symptom, while 30% (n=128) initially presented with an abdominal mass or swelling. The median duration of symptoms for the patients included in our study was 18 (5-24) months. In adolescents, the duration of symptoms shows a statistically significant positive correlation with tumor size (p=0.035, r=0.392) and with lesion size (p=0.039, r=0.346). No significant correlation was found between the duration of symptoms and cyst size (p=0.486). In pre-adolescents, the duration of symptoms shows a statistically significant positive correlation with lesion size (p=0.006, r=0.621). No significant correlation was found between the duration of symptoms and cyst size (p=0.413), or tumor size (p=0.104). In all patients, the duration of symptoms shows a statistically significant positive correlation with lesion size (p<0.001, r=0.460). No significant correlation was found between the duration of symptoms and cyst size (p=0.557), or tumor size (p=0.408) (Table II).

The duration of symptoms shows a statistically significant positive correlation with lesion size (p<0.001, r=0.460). The correlation between these items is shown in Figure 1.

By considering the duration of the patient's symptoms (in months), we can estimate the size of the patient's cyst/mass (lesion size). For this purpose, a model has been developed. The formula is a mathematical model developed to predict the size of a lesion (a cyst or mass) in centimeters, based on the duration of the patient's symptoms in months (Figure 2).

The cut-off value for laparotomy was calculated when compared with the group who underwent laparoscopy. The ROC analysis revealed different cut-off values for cyst, mass, and lesion sizes. The optimal cut-off value for cyst size was determined to be 5.2 cm, with an Area Under the Curve (AUC) of 0.62 (95% CI: 0.55-0.71). This cut-off demonstrated a sensitivity of 76% and a specificity of 36%. For mass size, the optimal cut-off was found to be 4.5 cm. This value yielded a higher AUC of 0.85 (95% CI: 0.74-0.96), with a sensitivity of 94% and a specificity of 55%. The lesion size cut-off was 5.7 cm, with an AUC of 0.66 (95% CI: 0.60-0.72). This value showed a sensitivity of 78% and a specificity of 36% (Table III).

The ROC (Receiver Operating Characteristic) curve of the mass size for laparotomy, which has the highest AUC (Area Under the Curve), is presented in Figure 3.

Table II. Correlation table (symptoms duration *vs*. lesions features).

			Age	Cyst size	Tumor size	Lesion size
Adolescent	Symptoms duration (hour)	*r*	-0.111	0.392	-0.076	0.346
		p	0.486	0.035	0.871	0.039
Pre-adolescent	Symptoms duration (hour)	*r*	-0.155	-0.492	0.668	0.621
		p	0.413	0.104	0.147	0.006
All-patients	Symptoms duration (hour)	*r*	-0.07	0.254	0.251	0.460
		p	0.557	0.110	0.408	<0.001

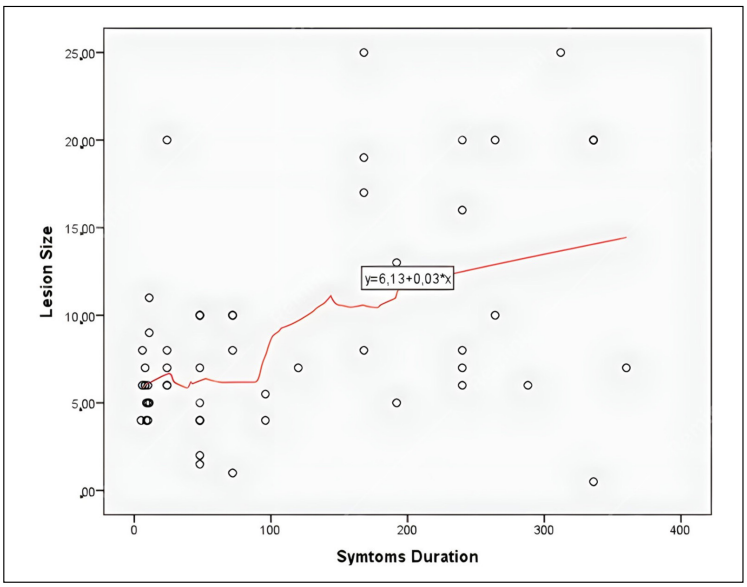

Figure 1. Correlation (symptoms duration *vs.* lesion size).

$$\text{Lesion size (cm)} = \frac{[613 + (\text{Symptoms duration X } 3)]}{100}$$

Figure 2. Lesion size / Symptom duration index.

Table III. The cut-off value and AUC analysis for laparotomy.

	Cut-off value	AUC	Sensitivity	Specificity
Cyst size	5.2 cm	0.62 (0.55-0.71)	76%	36%
Mass size	4.5 cm	0.85 (0.74-0.96)	94%	55%
Lesion size	5.7 cm	0.66 (0.60-0.72)	78%	36%

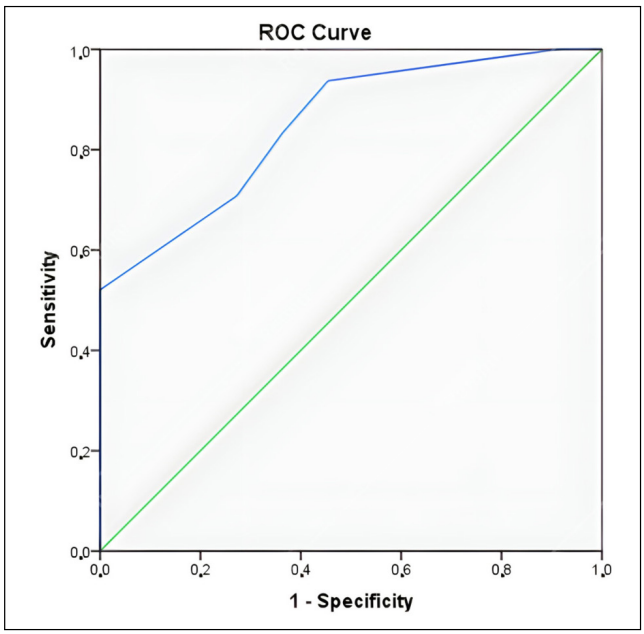

Figure 3. ROC analysis (mass size for laparotomy).

Discussion

Pediatric adnexal lesions manifest in various forms, including non-functional neoplastic ovarian cysts, ovarian torsion, benign tumors, and high-grade neoplastic diseases, with an occurrence rate of approximately 2-5 per 100,000[2,7]. These adnexal pathologies differ from adults in terms of the clinical picture they present, histopathological characteristics, diagnosis, and treatment methods. In children and adolescents, the pelvic region is situated higher and closer to the midline, and adnexal structures are positionally located higher than in adults. This leads to early symptoms and diagnosis of pathologies in this region[8]. Due to this anatomical structure, there is limited space for the development of adnexal masses, so complaints of abdominal pain, vomiting, and abdominal mass are more prominent than pelvic complaints. These masses often become symptomatic when a complication such as torsion, hemorrhage, rupture, or precocious puberty develops[8,9].

Acute symptoms usually arise due to torsion or rupture of a large cyst, resulting in associated bleeding[8]. Of the patients in our study, 65% (278 individuals) primarily reported abdominal pain as their initial symptom, while 30% (128 patients) initially presented with an abdominal mass or swelling.

This is the first study that estimates the size of the patient's cyst/mass (lesion size). For this purpose, we have developed a model. The formula is a mathematical model developed to predict the size of a lesion (a cyst or mass) in centimeters, based on the duration of the patient's symptoms in months.

The median duration of symptoms for the patients included in our study was 18 (5-24) months. Less commonly, patients may show signs of early puberty or virilization[10]. Depending on the clinical picture they present and their diagnosis, the treatment for these lesions can vary, ranging from conservative observation to medical treatment or surgical intervention. In the study conducted by Spinelli et al[11], surgery was performed on 40% of the patients. In contrast, in our study, the surgical incidence was 45%. Only those patients who underwent surgery were included in our study.

Our study's findings largely align with existing literature on pediatric adnexal pathologies. Regarding the etiology of adnexal pathologies, our study found cysts to be the most common, accounting for 67.1% of cases. This is consistent with the study by Cass et al[12] who also found ovarian cysts to be the most common adnexal mass in children and adolescents.

Our study indicates a preference for laparoscopic surgical interventions in the treatment of ovarian torsion, with 66.8% of the patient population undergoing this procedure. This contrasts with the literature, where the overall reported use of laparoscopy in these cases is only 23.5%[13]. However, this finding is not necessarily contradictory, as it can reflect a trend toward minimally invasive surgical practices over time. The high rate of laparoscopic surgery in our study aligns with the current recommendations stating that if technically feasible, laparoscopy should be the surgical approach of choice for ovarian torsion[14-17]. The advantages of laparoscopy include its role in reducing diagnostic delay and enhancing the feasibility of conservative surgery. This approach can be particularly valuable when the diagnosis remains unclear, thereby warranting initial intervention[14,15]. Garliner et al[13] provide a useful guideline suggesting that laparoscopy should be the chosen surgical approach for torsed ovaries measuring less than 75 mm in diameter, mainly due to the benign nature of the tumor at this size. Conversely, laparotomy should be performed for larger-sized tumors. In thus study, it would be informative to present the size distribution of the ovarian lesions among the patients who underwent laparoscopy *vs.* those who had a laparotomy. The cut-off value for laparotomy was calculated when compared with the group who underwent laparoscopy. The optimal cut-off was found to be 4.5 cm. This value yielded a higher AUC of 0.85 (95% CI: 0.74-0.96), with a sensitivity of 94% and a specificity of 55%.

Limitations

As the limitations, the study's retrospective design might affect the preciseness and completeness of the gathered data due to potential recall bias or missing medical records. The research was conducted at only two Turkish institutions, so its results might not apply universally due to variations in local healthcare practices and population attributes. The lack of follow-up data leaves long-term outcomes and complications unexamined. Additionally, without an evaluation of the pathology results, our understanding of the exact nature and diagnostic accuracy of the adnexal pathologies is hindered.

Conclusions

This study on adnexal pathologies in children indicates a shift towards minimally invasive laparoscopic surgery. A novel model predicting lesion size based on symptom duration was developed, and surgical intervention thresholds were established. This contributes to improved decision-making and enhanced fertility preservation in pediatric patients. Despite the positive findings of this study on pediatric adnexal pathologies, further investigation is required to validate the predictive model in other populations and evaluate its long-term outcomes.

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Conflict of Interest
The author has no conflict of interest to declare.

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Ethics Statement
The ethics approval for this study was obtained from the non-interventional local ethics committee of Başakşehir Çam and Sakura City Hospital (number: KAEK/2023.05.191).

Informed Consent
Informed consent was obtained from all patients.

Data Availability
Data are available on request from the authors.

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