ADOLESCENT OVARIAN TUMORS: A CLINICOPATHLOGICAL REVIEW OF 15 CASES

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Background: To prospectively review the clinicopathologic pattern and out come in 15 patients with adolescent ovarian tumours in Military Hospital Rawalpindi between Jan 2004 to Sep 2007. **Methods:** All cases of adolescent ovarian tumours reporting to the gynaecology department of Military Hospital Rawalpindi during the period of enrolment. Out of 15 cases 12 were managed surgically. Data was recorded regarding clinical presentation, patient's age, size of tumour, bilaterality, histopathology, staging if tumour was malignant and sites of extra ovarian involvement. All patients were followed up for one year. **Results:** Majority of patients fell in the subgroup 14–16 year age. Majority harboring ovarian malignancy belonged to subgroup 17–19 years. Clinical presentation in the majority was mass abdomen and abdominal distension. Approach was transcutaneous in 3 (20%), laparoscopic in 2(20%), and open laparotomy in 10 (60%). Frozen section was performed in 3 cases. Histopathology was benign in 11 cases and malignant in 4. In 3 cases cyst fluid was negative for malignancy. In 2 cyst wall biopsy and one patient where entire cyst was removed histopathology revealed benign serous cyst adenoma/luteal cyst (26%). There were 5 cases of dermoid cysts (33.3%) , one patient had bilateral dermoids, one malignant and one benign. All 4 malignancies were found to be to non-epithelial on histopathology. One case had surgery twice. Unilateral oopherectomy followed by recurrence and total abdominal hysterectomy and contralateral salpingo-oopherectomy after 2 years. **Conclusion:** This study shows the preponderance of non-epithelial tumours and high percentages of malignant germ cell tumours in adolescents. The incidence of malignant tumours in adolescents is higher than in adults. Early correct diagnosis could be reached by careful physical examination, imaging and tumour markers Surgery should as much as is safely possible be fertility preserving.

Key words: Ovarian tumours, Adolescent, Fertility preservation

INTRODUCTION

Ovarian tumours occurring in adolescent age group are unique with regard to their rarity, notoriously lethal when malignant and controversial in management. Due to infrequent occurrence most gynaecologists are not familiar with special problems inherent with these neoplasms.[1] The surgical and medical management in adolescents can be modified when feasible and safe in order to maintain patient's reproductive and menstrual capabilities. It is important that gynaecologists should remember the differential diagnosis and treatment options for adnexal masses, both so that significant pathology can be treated surgically when indicated and also expectant management can be offered when appropriate to avoid potentially unnecessary surgery with its inherent risks and sequelae.

Ovarian tumours are rare in childhood and the adolescents and are reported to be 2% of all the cases seen.[2] If present there is a 25% chance that the tumour is malignant. These tumours are rare accounting for 1% of malignant neoplasm in this age group; ovarian tumours occur in young girls and can be discovered due to symptoms, on physical examination and or through imaging studies.[1]

An ovarian mass that is purely cystic and no complex features is almost certainly benign and can be managed by observation. One study that expectantly managed 51 girls with simple and complex cysts reported that 90% of cysts resolved spontaneously.[2] Ovarian masses presenting with torsion are usually benign.[3] Asymptomatic are more likely to harbor a malignancy. Young ladies between menarche and 19 years constitute this age group. In this group simple and complex cysts are both relatively common. Most simple cysts result from failure of maturing follicle to ovulate and involute.

Most ovarian cysts in perimenarchal period are asymptomatic, but may cause menstrual irregularities, pelvic pain or pressure symptoms. Asymptomatic simple unilocular cysts 7–10 cm on ultrasound examination can be observed with or without oral contraceptive pills. Patient should be evaluated monthly or fortnightly. The challenges facing the gynaecologist are carelessness leading to under treatment and under quality of care and extreme surgical intervention leading to over treatment.

Internationally data on teenage ovarian tumours is scarce. Local studies are also deficient. Aim of our study was to see the pattern and outcome of disease in our population.

MATERIAL AND METHODS

All cases of ovarian tumours reporting to the gynaecology department of Military Hospital (MH) Rawalpindi during the period of enrolment were collected and analyzed. Except 3 out of the 15 cases all were managed surgically. Data was recorded regarding clinical presentation, patients' age, size of tumour,

bilaterality, type of surgery, staging if tumour was malignant and sites of extra ovarian involvement, histopathology and chemotherapy. All patients were followed up for at least one year.

RESULTS

Majority of patients belonged to the age group 14–16 years (Table-1). In this study 4 cases were malignant and 3 out of these four cases belonged to 17–19 years age group Clinical presentation is shown in Table-2. Five (33.3%) patients presented with mass and abdominal distension and a similar number presented with mass and pain. Incidental diagnosis during workup of menstrual irregularity was made in 3 (20%) patients.

Ultrasound revealed cystic tumours in 7 patients (43.75%). Solid tumours in 4 cases (25%) and 5 patients (31.25) had typical sonographic appearance of dermoid (Table-3). Three tumour markers were done in all cases: Ca-125, b-HCG and alpha-fetoprotein. In only one case of solid ovarian tumour b-HCG was raised and the tumour turned out to be dysgerminoma.

Surgical approach was transcutaneous in 3 patients (18.75%) as shown in Table-4. In both these cases cysts were 8-10cm in diameter and very thin walled. Both patients belonged to highly educated family and were expected to be compliant at follow up.

Two patients were subjected to laparoscopy with cyst aspiration and cyst wall biopsy. In both these cases cysts were 10–12 cm and unilocular. In 11 patients (68.75%) tumour was approached by laparotomy, that was paramedian in 5 (one patient had 2nd laparotomy through same approach) and pfenniestiel in the remaining 5. Paramedian approach was chosen in all solid looking ovarian tumours (all turned out to be malignant) and one large thick walled mucinous cystadenoma size of a 28 week pregnancy.

Cystectomy was done in 6 patients (37.5%), 5 had dermoids and one had large simple cyst about 18 cm. (Table-5). In all these cases ovarian tissue was conserved and reconstructed. Frozen section was arranged in 5 patients but was only performed in 3 individuals. The other 2 tumours were confined completely to one ovary and frozen section would not have changed the conservative management plan. Unilateral oopherectomy was done in 3 (18.75%). Two of malignancy and also one large thick walled cyst. Total abdominal hysterectomy was done as a primary procedure in the young lady with bilateral ovarian involvement, ruptured capsule, massive ascites and sudden onset of virilization with Sertoli Leydig cell tumour. One unfortunate patient under went laparotomy twice. Once at the age of 11 years for unilateral dysgerminoma followed by chemotherapy. Unfortunately she had recurrence after 2 years. Total abdominal hysterectomy with contralateral salpingo-oopherectomy was done after ureteric stenting as there was right sided hydronephrosis due to secondaries in the pelvis. We tried to save fertility in three out of four patients but managed to save it in only half the cases at the end.

In the 10 cases that underwent laparotomy, histopathology showed 5 (31.25%) of dermoids, 2 (12.5%) dysgerminoma, one (6.25%) malignant dermoid and one serous cystadenoma (Table-6). All case were followed up for at least a year

Table-1: Age at presentation (n=15)

Age	Number	%
11–13 years	3	20.0
14–16 years	7	46.7
17–19 years	5	33.3

Table-2: Clinical presentation (n=15)

Presentation	Number	%
Mass with abdominal distention	5	33.3
Mass with pain abdomen	5	33.3
Incidental	3	20.0
Pelvic pain	2	13.3

Table-3: Ultrasound appearance (n=16, one patient had recurrent tumour after 2 years)

Appearance	Number	%
Cystic	7	43.75
Solid	4	25.0
Typical dermoid	5	31.25

Table-4: Surgical approach (n=16, one patient had laparotomy twice)

Approach	Number	%
Transcutaneous aspiration	3	18.75
Laparoscopy	2	13.3
Laparotomy	11	68.75

Table-5: Surgical outcome (n=16, one patient operated twice and one had bilateral tumours)

Operation	Number	%
Cystectomy***	6	37.5
Unilateral oopherectomy**	3	18.75
Radical surgery*	2	13.3
Aspiration with biopsy	2	13.3
Aspiration only	3	18.75

*Includes Total abdominal hysterectomy with bilateral salpingo-oopherectomy, omentectomy and debulking.
** In one case aortic lymphadenectomy and jejunostomy also done.
***One patient had ipsilateral cystectomy with contralateral salpingo-oopherectomy

Table-6: Histopathology. (n=16, one patient had bilateral tumours)*

Type	Number	%
Simple/serous ovarian cyst	7	43.75
Dermoid cyst	5	31.25
Dysgerminoma	2	12.5
Sertoli Leydig cell tumour	1	6.25
Malignant dermoid	1	6.25

DISCUSSION

Treatment of ovarian tumours in adolescent generally adheres to the principles of adult management, with more emphasis on anatomic preservation.

Ovarian tumours account for approximately 1% of all tumours in children and adolescents. Less than 5% of ovarian malignancies occur in this age group. 30 % of all ovarian neoplasm occurring during childhood and adolescence are malignant.[4] Although gynecologic tumours are infrequently seen in childhood and adolescence, they should be included in the differential diagnosis of mass abdomen. The incidence of malignant tumours is higher than in adult Germ cell tumours and make up to half to one third of ovarian neoplasm in girls up to 19 years of age in the study by Shultz K *et al.*[5] In our study 60% of tumours were non-epithelial. Unlike the world data where malignant ovarian tumours peak between 11–13 years in our study the peak was at 17–19 years.[6] Benign dermoid were uniformly distributed in all age brackets in our study. Clinical presentation is somewhat different than the adults. Symptoms may not be associated with the lesion and sometimes ovarian enlargement may be big enough so as to produce a pelvic swelling or tumor that can be felt in the abdomen in the pre pubertal and pubertal age group.[2] In an Indian study, pain was the most frequent symptom followed by mass. In the present study mass and pain/mass with abdominal distension were the most common presentation that led to diagnosis. In Lind fort's series pain was the main presenting complaint followed by abdominal distension and torsion[8] which was different from our study. Menstrual irregularities and weight loss was the next presentation in Lindforte's[8] series which was again at variance with our study. In a review of 81 cases from Lindforte's 35% of cases were malignant. Huffman[9] reported that 30% of 999 tumours and Breen and Maxson 27% of 1309 tumours in children and adolescents were malignant.[2,5] In another series from Nepal malignancy rates have varied from 15–35%. In our study 26% of cases were malignant. Like international data germ cell tumours and sex cord stromal tumours were preponderate in adolescents.[2,3] In agreement with our study dysgerminoma account for 5–10% in the age group below 20 years of age.[6]

In this study, 74%% ovarian tumours were benign and 26% were malignant. This is similar to the data from western countries where 75–80% of ovarian tumours are benign.[1] Also study carried in India by Pilli *et al*[7] had approximately similar results which showed that 75.2% ovarian tumours were benign .However this figure was only 59.2% in study carried in Pakistan by Ahmad *et al*.[10] Surprisingly results of two studies from India are different.[7,10] This study as well as studies from India and Pakistan which are included here for comparison have small sample size and are institution based where as most data of western world is taken from results of large population based studies. In study of Hassan *et al*[12] in 1st two decades, 49.1% tumours were germ cell tumors similar to the results in our study. In this study, under 21 years of age 11 ovarian tumours were seen, out of which 8 (72.7%) were germ cell tumors. 100.0% of malignancies in 1st two decades were non-epithelial in origin. We also had identical results did not have a single case of epithelial ovarian carcinoma. This study shows the preponderance of germ cell tumour and high percentages of malignancies within germ cell tumour in young children and adolescents This study as well as studies from India and Pakistan which are included here for comparison have small sample size and are institution based, where as most data of western world is taken from results of large population based studies[1]. This could be one cause of variation in results. Our study is also institution based and has small sample size. So the result obtained may or may not reflect the actual histological pattern and age distribution of ovarian tumors in young women in Pakistan. There is need for bigger population studies with larger sample size.

In smaller thin walled cysts fluid aspiration in 3 patients was not associated with recurrence. These are in agreement with study by Lipitz *et al*.[13] For fertility preservation laparoscopic approach should be adopted where possible .This was demonstrated in the study by Pansky M *et al*, where premenarchal girls were managed by laparoscopic ovarian cystectomy.[14,15] Benign ovarian cysts and paratubal cysts are often amenable to cystectomy, allowing preservation of the remaining ovarian cortex. This often can be done, even with large cysts with little or no normal obvious ovarian tissue visible. There is risk of intraoperative spillage of cyst contents during manipulation with cystectomy, and this is problematic if the cyst exhibits borderline or frank malignancy, or if a dermoid spills with its risk of peritonitis and subsequent adhesion formation. We managed 2 teenagers laparoscopically. Unfortunately one was lost to follow up.

Imaging in ovarian tumours is done by ultrasongraphy .It gives information. Dermoids can present with dull abdominal pain but were frequently asymptomatic and often found by exam or incidental imaging. They have a characteristic ultrasound appearance with fat fluid levels, diffuse or focal areas of increased echogenicity with acoustical shadowing, often thought to be hair fibers within the cyst, and may contain a mural hyperechoic nodule. In solid ovarian tumours we also performed computerized tomography to assess stage of the tumour preoperatively. Doppler ultrasound has been found to be helpful in cases suspicious of malignancy but was not used in our study.

Gentle tissue handling, meticulous hemostasis and adhesion prevention strategies were adopted for fertility preservation. Frozen section was performed in 3 cases, though it was arranged in 5 cases. In two cases we did not use the facility as it was not going to effect our management in localized tumours. In case of bilateral dermoid one obviously benign (with vernix and

hair) and the other suspicious and solid looking, we tried to conserve both ovaries. When the result of frozen section confirmed malignancy we had to sacrifice one ovary in the 17 year old girl. Frozen section also confirmed malignancy in 18 ½ year old recently married girl with bilateral Sertoli Leyig cell tumour and massive ascites. The family was deeply grieved but gave consent for total abdominal hysterectomy and bilateral salpingo-oopherectomy. She is now in complete remission for the past 3 years. Voice changes and hirsuitism still persist. Literature also shows that 70.0% metastatic ovariantumors are bilateral.[12]

In one case of bilateral dysgerminoma with secondaries in aortic lymph nodes and mass involving duodenal cap, conservative surgery was done. Ipsilateral salpingo-oopherectomy and jujenostomy was done. She is at the moment in complete remission 1½ year following chemotherapy with Bleomycin, Etoposide and Cisplatin These chemotherapeutic agents do not interfere with fertility and there have been no long term reports of congenital abnormalities in children born to mothers who received this chemotherapy.[3] The overall survival and cure rate of patient with non epithelial malignancy has improved dramatically in adolescents with chemotherapy. Surgical intervention is directed towards preservation of reproductive function .Unless malignancy is diagnosed definitely at the time of surgery on frozen section conservative surgery should be undertaken. It is preferable to subject the patient to second procedure after the final pathological specimens are reviewed than to perform unnecessary ablative procedure.[16] In addition, with advances in assisted reproductive technologies, it is currently possible to achieve pregnancy using donor eggs following bilateral oopherectomy, or to donate eggs for surrogate pregnancy following hysterectomy, so only the necessary pelvic organs should be removed if indicated. Support and sensitivity for the frightened girl and her family is essential as is high quality decision making by the gynaecologist.

The incidence of malignant of tumours in adolescents is higher than in adults. Therefore, the detection of these lesions is worrisome to patients, her families, and gynaecologists. Early correct diagnoses and preservation of fertility with should be the aim

CONCLUSION

Adolescent ovarian tumours though rare are important as any solid ovarian tumour in this age group should be considered malignant until proved otherwise by histological examination. For benign tumours adhesion prevention strategies should be used. Surgical intervention should as much as possible be directed towards preservation of ovarian tissue. There is scarcity of published literature on this subject. We need bigger studies to address the issue of how much fertility preservation is safely possible. Irrespective of indication for surgery, it is always preferable to attempt conservative, fertility-sparing surgery in adolescents. It is desirable to embark upon a second surgery if final pathology reveals a malignancy, rather than proceed with empiric extirpative surgery.

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