Case Report

A huge ovarian mucinous cystadenoma associated with contralateral teratoma and polycystic ovary syndrome in an obese adolescent girl

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A 13-year-old, obese girl presented with acute abdominal pain with abdominal distension for a year. The physical examination revealed marked abdominal distension with a large well-circumscribed mass sized 13×20 cm. Her body mass index (BMI) was 37.8 kg/m^2. An abdominal CT scan revealed a huge multiloculated cystic mass and a left adnexal mass. She had an abnormal fasting plasma glucose and low HDL-C. Laparotomy, right salpingo-oophorectomy, left cystectomy, lymph node biopsies and partial omentectomy were performed. The left ovary demonstrated multiple cystic follicles over the cortex. The histologic diagnosis was a mucinous cystadenoma of the right ovary and a matured cystic teratoma of the left ovary. Both obesity and polycystic ovary syndrome (PCOS) are associated with a greater risk of ovarian tumours, where PCOS could be either the cause or as a consequence of an ovarian tumour. We report an obese, perimenarchal girl with bilateral ovarian tumours coexistent with a polycystic ovary and the metabolic syndrome.

Key Words: epithelial ovarian tumour, teratoma, polycystic ovary syndrome, obesity, adolescent

INTRODUCTION

Ovarian tumours in children are uncommon and represent less than 2% of all tumours in girls younger than 16 years.[1] Bilateral ovarian tumours are even much rarer in this age group. Polycystic ovary syndrome (PCOS) was defined at a meeting of experts in Rotterdam, 2003, to require two of the three following features: i) clinical or hormonal evidence of androgen excess; ii) oligomenorrhoea or amenorrhoea; iii) ultrasonographic evidence of polycystic ovaries.[2] PCOS is hypothesized to have some roles in the pathogenesis of the ovarian tumours. We present a case of bilateral ovarian tumours coexistent with polycystic ovary in an obese perimenarchal girl.

CASE

A 13-year-old girl presented with acute abdominal pain for 4 days. She had abdominal distension for a year without any gastrointestinal symptoms. She had menarche since a year ago with irregular cycle. She had dull abdominal pain during her period for 4 days. She was obese from the age of 5 years. Her mother, grandmother and grandfather all had diabetes. The physical examination revealed marked abdominal distension with a large well-circumscribe, firm mass sized 13×20 cm with an irregular surface and mild tenderness. Her body weight was 103 kg, height 165 cm and body mass index (BMI) 37.8 kg/m^2. An abdominal CT scan revealed a huge multi-loculated cystic mass, sized 20×30×33 cm with thin septation without calcification which occupied nearly the entire abdominal cavity. There is also left adnexal fatty mass 6×6×6 cm with internal soft tissue strands without calcification. The patient's complete blood count, urinaysis, renal and liver functions were normal. She had an abnormal metabolic profile as follows: total cholesterol 4.66 mmol/L, LDL-C 3 mmol/L, HDL-C 1.01 mmol/L, triglyceride 1.07 mmol/L, fasting plasma glucose 5.89, 1-hr 75 g OGTT 10.1 and 2-hr 7.39 mmol/L. Tumour markers were as followed: β-HCG <0.1 mU/mL, α-fetoprotein 0.79 mcg/L, CA-125 112 kU/L. Laparotomy was performed through midline incision. Clear yellowish peritoneal fluid, approximately 250 mL, was noted. There was a huge, one-and-a-half round twisted right ovarian cyst, sized 50×30×40 cm with omental attachment. A 5-cm cystic mass of the left ovary was seen (Figure 1). The left ovary appeared to have multiple cystic follicles over the cortex. The omentum, peritoneum, liver, pelvis and uterus were all normal. Right salpingo-oophorectomy, left cystectomy, lymph node biopsies and partial omentectomy were performed.

Macroscopic examination of the right adnexal mass re-

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Figure 1. Operative findings of both ovarian tumours. (A, B) Right ovarian cyst, (C) Left ovarian cyst.

vealed a grayish, cystic mass with smooth surface. A cross-section showed a multiloculated cyst containing mucous without a solid part. The left ovarian cyst contained yellowish oily material and hair with grayish-white tubercles. Histopathologic examination of right ovarian adnexa revealed cystic structures lined with mucinous epithelium with an unremarkable tube. No metastatic findings of the pelvic nodes and omentum were found. The final histologic diagnosis was mucinous cystadenoma of the right ovary and matured cystic teratoma of the left ovary.

SUMMARY AND CONCLUSION

Primary ovarian tumours are relatively uncommon in childhood. The incidence of ovarian tumours has been estimated to be less than 2% of all tumours in girls less than 16 years of age.[1] Epithelial ovarian tumours represent 13-19% of all ovarian neoplasms in childhood.[3,4] The majority of cases presented in adulthood suggest that ovarian epithelial tumours may be stimulated by hormones. The most common type of epithelial tumours in children is a benign cystadenoma[3,5] and the serous type is more common than the mucinous type.[4,6] Mucinous ovarian tumours, as presented in this girl, mostly occur in middle adult life and are extremely rare in the perimenarchal period.[1] In contrast, teratoma are the most common germ cell tumours of childhood ovarian tumors.[7] These tumours are mostly non-secreting. There are some reports of androgen-producing ovarian teratoma, which is relatively rare in childhood.[8]

This girl was also diagnosed as metabolic syndrome: abdominal obesity, abnormal fasting plasma glucose and low HDL-C; and polycystic ovary syndrome: the features of oligomenorrhea, hirsutism and polycystic ovary were seen in operative findings. Obesity and metabolic syndrome seems to play an important role in the development of PCOS. As compared with normal and underweight girls, the odds ratios for PCOS diagnosis were 3.85 (95% CI: 3.04-4.88), 10.3 (95% CI: 8.16-12.8), and 23.1 (95% CI: 18.7-28.6) for overweight, moderately obese, and extremely obese adolescents, respectively.[9] The prevalence of metabolic syndrome among adolescents with PCOS varies from 11.8-33.3%,[10-12] determined partly by race, ethnicity and the criteria used for making the diagnosis.

Both obesity and PCOS are associated with a higher risk of ovarian tumour. The risk of ovarian cancer increases with increasing BMI. A systematic review of epithelial ovarian cancer demonstrated that overweight or obesity in early adulthood is associated with an increase risk of ovarian cancer with the estimate risk of 1.22 (95% CI: 1.02-1.45).[13] The positive association among ovarian cancer and BMI is stronger with the BMI at age 18 as compared to current adult BMI.[14] There is no current evidence that the association varies among different subtypes of ovarian cancer. These findings suggest that adolescent obesity is associated with gynecological cancer risk and the physicians should be aware of this.

In our case, this perimenarchal girl has bilateral ovarian tumours coexistent with polycystic ovary. PCOS could be either the cause or the consequence of ovarian tumours. There are some studies that reported the coexistence of PCOS and androgen-producing ovarian teratoma.[8,15] Contralateral epithelial ovarian tumour and PCOS are associated with a high frequency of hyperplastic and metaplastic changes in the surface epithelium in ovaries as compared to patients without disease as well.[16,17] These imply the possible increase in ovarian cancer risk among women with PCOS, which could support the gonadotropin, androgen, progesterone or insulin hypotheses. Although the role of PCOS in ovarian tumour is not yet established, the elevation of serum androgen and LH, as the cardinal features of PCOS, appears to be involved in ovarian carcinogenesis.[18,19] In addition to long term cardiovascular and metabolic complications of PCOS, ovarian tumours are not unusual in women with PCOS and need to have the regular follow-up.

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AUTHOR DISCLOSURES

All authors declare that there are no conflicts of interest. This study was approved by the ethical committee of the Faculty of Medicine of Thammasat University.

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肥胖少女巨大卵巢粘液性囊腺瘤与对侧畸胎瘤和多囊卵巢综合症有关

一个 13 岁肥胖女孩腹胀一年，急性腹痛入院。体格检查发现明显腹胀，有一个尺寸为 13×20 cm 边界清楚的肿块。她的体质指数为 37.8 kg/m^2。腹部 CT 扫描发现一个巨大的多腔囊性肿块和左侧附件有一包块。她的空腹血糖异常，HDL-C 低。进行剖腹探查，右侧输卵管卵巢切除术，左囊切除术，淋巴结活检和部分网膜切除术。左侧卵巢皮质发现多个囊性卵泡。组织学诊断为右侧卵巢粘液性囊腺瘤和左侧卵巢成熟囊性畸胎瘤。肥胖和多囊卵巢综合症与卵巢肿瘤高风险有关，而多囊卵巢综合征可能是卵巢肿瘤的原因或是结果。我们报告了一个肥胖、围初潮期女孩，双侧卵巢肿瘤合并多囊卵巢综合征和代谢综合征。

关键词：卵巢上皮肿瘤、畸胎瘤、多囊卵巢综合征、肥胖、青少年