Case Report

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Bilateral ovarian tumour in a young girl

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ABSTRACT

Bilateral ovarian tumour in a girl presents the dilemma of conservative versus aggressive approach towards these tumours. When faced with suspicious tumour and complete replacement of the ovaries bilaterally, bilateral oophorectomy is a viable option, though the certain possibility of infertility and lifelong hormonal supplementation is unavoidable. We report a case of bilateral ovarian masses in a young girl, which on histopathological examination showed mature teratoma with aggregates of proliferating capillary and cavernous sized vessels in the tumour wall. Such associations are rare and must be differentiated from a vascular neoplasm.

Key words: Bilateral ovarian tumour, mature cystic teratoma, vascular proliferation

INTRODUCTION

Teratomas are the most common benign tumour of the ovary.[1] Mature cystic teratomas have classical morphological features. The occurrence of vascular proliferation in a mature teratoma is uncommon, though many morphological variations have been quoted in the literature. There have been only a few case reports on the occurrence of florid vascular proliferation and haemangiomas in teratomas.[2-10] Also, the peculiarity of bilateral ovarian teratomas in a child is a rarity meriting report of the case.

CASE REPORT

A 3-year-old child presented with pain abdomen and abdominal distension for 3 weeks. On examination, bilateral firm nontender, ballotable loin masses were palpable. The parents denied history of malignancy in the family. Sonographic imaging showed bilateral variegated masses arising in the pelvis, suggestive of ovarian tumours. Computed tomography (CT) contrast demonstrated the vascularity of the mass. In the workup for a germ cell tumour, alpha-fetoprotein and beta human chorionic gonadotrpin (βhCG) levels were found to be normal. Child was subjected to surgery after a detailed informed consent was taken. At laparotomy, prominent vessels feeding the tumours were noted without ascites, peritoneal deposits and para-aortic lymphadenopathy. Bilateral oophorectomy was performed with partial omentectomy and nodal sampling. At follow-up of 6 months post surgery, the child was well without clinical and sonological recurrence.

Pathology

Gross examination showed bilaterally enlarged ovaries (right 12.5 × 7 × 2 cm, left 5.5 × 5 × 2.5 cm) with a variegated appearance having solid and cystic areas. The solid areas appeared grey brown, congested and spongy. There were areas with cysts containing cheesy white pultaceous material [Figure 1]. Further cut sections of the mass revealed a focus showing mucinous material.

Microscopic examination showed a solid cystic structure lined by keratinised stratified squamous epithelium with fibrocollagenous wall showing florid proliferation of both closely packed cavernous [Figure 2] and capillary sized vascular channels. Multiple sections did not show any immature component. Only on further careful sampling from the grossly appearing mucinous areas, occasional ciliated columnar epithelium-lined glands (airway-like) were seen intermingled with the vascular proliferation. The epithelium also showed crypt-like architecture in some foci; however, goblet cells were not seen. Immunohistochemical staining with CD31 highlighted the vascular spaces. No normal ovarian parenchyma was seen. Omental sampling was histologically

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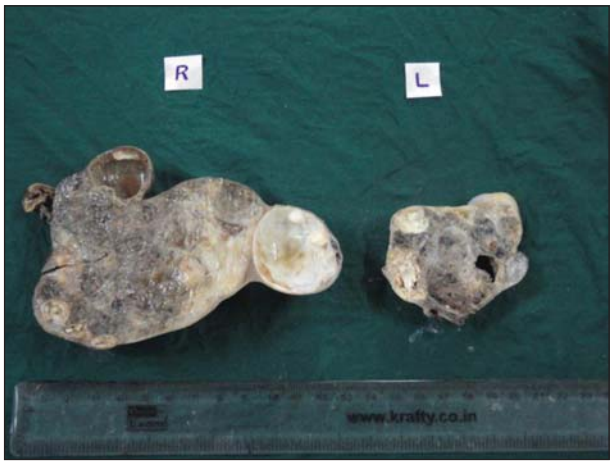


Figure 1: Cut surfaces of both ovaries showing solid cystic areas with some cysts containing cheesy white pultaceous material

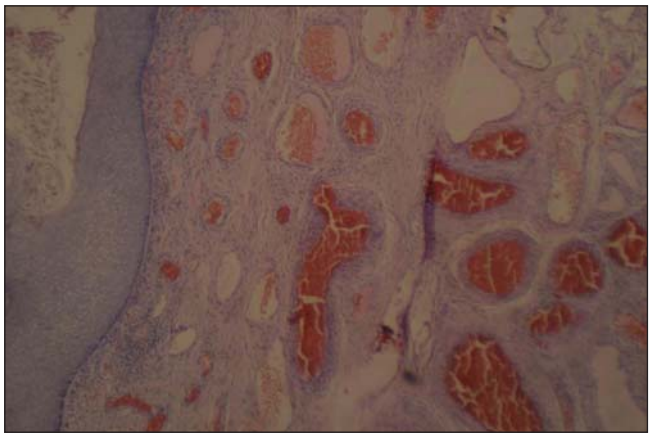


Figure 2: Photomicrograph showing cyst lined by stratified squamous epithelium containing keratin flakes and showing numerous capillary and cavernous channels in the wall (HandE, ×40)

unremarkable and section from the mesenteric lymph node showed reactive hyperplasia.

DISCUSSION

Mature cystic teratoma comprises 25% of all the ovarian tumours and is the most common benign tumour of the ovary. It is bilateral in 10-15% of the patients. These tumours usually develop in children or women in the reproductive age group. They contain cells derived from any of the three germ cell components — ectoderm, endoderm and mesoderm.[1]

The various tissue components seen in a teratoma include skin, cartilage, smooth muscle, respiratory epithelium, intestinal mucosa, neural epithelium, etc. However, the occurrence of florid vascular proliferations is rare in a teratoma, with only few case reports documented in the literature.[2,3,8-10] In the index case, stratified squamous epithelium with florid vascular proliferation was seen in the absence of any immature component. The importance of identifying these aggregates of small- and large-sized vessels as proliferations is to avoid a misdiagnosis of a vascular neoplasm. These cases should not be considered as immature teratomas either. There have been case reports of association of teratomas with haemangiomas.[4-6] In this case, after thorough sampling, columnar lined tissue was seen closely intermingled with the vascular proliferations, thus excluding the possibility of a vascular neoplasm.

In the case reports of vascular proliferation in teratomas, these proliferations have been found along with neural tissue.[3,7-10] Florid vascular proliferations have been noted in neural and neuroendocrine tumours.[9] It has been attributed to angiogenic factors released by the neural tissue. It has also been proposed that the mature tissues of the ovarian teratomas may produce angiogenic factors causing vascular proliferations in them, like the neural tumours. However, in this case, even after thorough sampling of the tumour, no neural tissue was present; rather, a mature columnar epithelial component was seen which could have been the contributing factor for the vascular proliferation.

Tatsuro *et al.*, in a recent series of paediatric ovarian tumours, noted bilaterality in 1.7% (1 out of 60), in whom ovarian preserving resection was done.[11] The recommendation of tumour resection with ovarian preservation could not be carried out in our case as the tumours appeared vascular during surgery with the suggestion of a possible malignancy, and further on histology, no demonstrable normal ovarian tissue was made out.

CONCLUSION

Though florid vascular proliferations are rare in teratomas, recognition of these as reactive non-neoplastic proliferations is essential to avoid a false diagnosis of vascular neoplasm or immature teratoma, and this requires thorough sampling of the tumour. Bilateral mature cystic teratomas in a child pose management challenges with regard to ovarian conservation.

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