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Pre- pubertal Yolk sac tumor: a Case Report

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ABSTRACT

Pediatric testicular tumors account for 1% to 2% of all pediatric tumors. These tumors are significantly different from their adult counterparts in terms of tumor histopathology, malignant potential and pattern of metastatic spread. The majority of prepubertal testicular tumors (children aged < 12 years) are yolk sac tumors followed by teratomas and stromal tumors. In 90% of these cases, AFP (α- fetoprotein) levels are elevated which is helpful in pre- operative distinction between yolk sac tumor and other tumors. This case report describes the clinical presentation, radiological findings, diagnosis and management of yolk sac tumor in an eighteen months old child.

Keywords: Yolk sac tumor, α- fetoprotein, Testicular tumor.

INTRODUCTION

Testicular germ cell tumors have a bimodal age distribution with one peak occurring during the first two years of life and a second peak occurring in young adulthood [1]. Among adults, seminomas and mixed germ cell tumors (MGCTs) account for majority of testicular tumors whereas majority of pediatric testicular tumors are yolk sac tumors. In prepubertal age (boys < 12years), seminomas or MGCTs are rare [2]. Testicular tumors typically present as painless solid masses. Ultrasound examination is an excellent tool to distinguish intratesticular from extratesticular masses such as epididymal cysts. AFP (α- fetoprotein) levels are elevated in most cases of yolk sac tumors. Preoperative AFP levels help in distinction between yolk sac tumor and other tumors. Post- operative AFP levels are an important surveillance parameter [3]. Approximately 80% of prepubertal patients with testicular tumors have clinical stage I disease and therefore can be managed with orchidectomy and observation [4]. We report a rare case of prepubertal yolk sac tumor in an eighteen-month-old child.

CASE REPORT

An eighteen months old boy was brought by his mother to the Department of Surgery with a painless swelling of left hemiscrotum for one month. There was no history of fever, trauma or urinary tract infection. The general physical examination was normal and the child had normal developmental milestones. On local examination, the left scrotal mass was non- tender, firm in consistency and measured 6.0x4.5x4cm in size. Ultrasound examination revealed significantly enlarged left testis (4.9x3.5x4.3cm with volume 39.8c.c). It showed heterogenous echotexture with multiple cystic areas in the parenchyma. MRI perineum showed well circumscribed, large heterogenous mass lesion replacing left testis. The mass revealed isointense signal intensity on T_1W images and heterogeneously

hyperintense signal intensity on T_2W/ STIR images with cystic areas within. The mass measured approximately 53 (CC)x 35 (AP)x 37(TR)mm. The right testis was normal in size, shape and signal intensity and normally located in scrotal sac. It measured approximately 12.7x 8.1mm. There was no evidence of any pelvic lymphadenopathy. All other pelvic tissues and organs were unremarkable. Chest radiograph was normal. All other routine laboratory investigations were within normal limits. The serum AFP level was found to be markedly elevated for age (1054.4ng/ ml) with normal beta human chorionic gonadotrophin (HCG) levels. The patient underwent unilateral high inguinal orchidectomy. The left testis was removed together with all its coverings with maximum cord length up to the deep inguinal ring. The specimen was submitted for histopathological examination. Grossly, the testis and spermatic cord measured 5.5x 4.5x 3.5cm and 4x1cm respectively. External surface was congested. The cut surface of testis showed a homogenous, grey white, circumscribed area measuring 4.5x 4.5cm. Few minutely cystic areas and pinhead size areas of hemorrhage were also seen. The histopathological examination revealed a tumor composed of epithelial and mesenchymal elements arranged in microcystic, glandular- alveolar, perivascular and papillary formations. At places, solid cords of cells were also seen. The cystic spaces were lined by flattened, endothelium like layer of cells. Perivascular Schiller-Duval bodies were also noted. Intracytoplasmic and extracytoplasmic eosinophilic inclusions were seen at places. Occasional areas of hemorrhage and necrosis were noted. The tumor cells were seen infiltrating the tunica albuginea but not invading through it. Microsections from the epididymis and spermatic cord were free from tumor infiltration. At six months follow- up, the patient was doing fine with latest AFP levels of 5.5ng/ ml.

DISCUSSION

Testicular tumors are far more common in adults than in children. Among adults, germ cell tumors account for 90% of testicular tumors; of which yolk sac tumors comprise less than 1% cases [2]. In children, germ cell tumors account for 60- 70% of testicular tumors [5]. Teratoma and yolk sac tumors are the most common type of germ cell tumors in children. Almost half of the pediatric testicular tumors are yolk sac tumors followed by teratomas and stromal tumors [6]. Several studies suggest that teratomas are more common than yolk sac tumors in pediatric patients; however, these studies were not limited to prepubertal patients [7- 9]. There are important differences between testicular tumors occurring in children and those occurring in adults. These involve tumor histopathology, malignant potential and pattern of metastatic spread. These differences are significant thus warranting a distinct approach to the treatment of prepubertal tumors. AFP is a reliable tumor marker which is elevated in 90% of patients with yolk sac tumor. But AFP levels are quite high in healthy infants. AFP levels are approximately 50,000ng/ ml in newborns, dropping to 10,000ng/ ml by age 6 months. So, AFP may be less helpful in distinguishing tumor types in young infants. The half- life of AFP is approximately 5 days. Failure of an elevated AFP to decline as expected after removal of primary tumor is attributed to persistent metastatic disease [2]. Approximately 80% of prepubertal patients with testis tumors have clinical stage I disease (tumor is confined to the testis) [4]. Invasion of epididymis, tunica albuginea, spermatic cord or scrotum does not change tumor stage but increases the risk of nodal involvement and the risk of recurrence [1]. These patients can be managed by orchidectomy and observation. The retroperitoneal lymph node dissection (RPLND) is not required for prepubertal testicular tumors because the pattern of metastatic spread is not same as seen in adults. Among pre- pubertal patients with metastatic disease, a minority have a disease limited to the retroperitoneum; the majority have disease in the chest (with or without retroperitoneal disease) [4]. The morbidity associated with abdominal surgery is greater for children than for adults. They have a high rate of complications like post- operative bowel obstruction, chylous ascites and subsequent ejaculatory dysfunction [2, 4]. The recurrence rate for the stage I patients managed with orchidectomy and observation alone varies from 15- 20%. These patients can be cured successfully by combination chemotherapy [4, 10]. In stage II, the tumor has retroperitoneal lymph node metastasis. Stage III is characterized by supraclavicular lymph nodes, visceral involvement or persistently elevated tumor marker levels. As this patient had stage I disease, so the management plan was high inguinal orchidectomy and strict follow up at least for two

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years. Follow up includes frequent chest and abdominal imaging and measurement of AFP levels.

CONCLUSION

There has been a dramatic improvement in prognosis of testicular tumors even in metastatic disease. Despite that prepubertal testicular tumors result in great anxiety and parents have reservations regarding the treatment, any recurrence and its effects on the child's growth and fertility. The prepubertal testicular tumors are distinct from those seen in adults in several aspects. Most of these tumors present in stage 1 disease and thus can be managed with orchidectomy and strict surveillance.

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FIGURES

Fig 1a: Left scrotal mass measuring 6.0x4.5x4cm in size; **1b:** USG examination revealed significantly enlarged left testis (4.9x3.5x4.3cm with volume 39.8c.c). It showed heterogenous echotexture with multiple cystic areas in the parenchyma; **1c:** MRI perineum showed well circumscribed, large heterogenous mass lesion replacing left testis. The mass measured approximately 53 (CC)x 35 (AP)x 37(TR)mm; **1d:** The patient underwent unilateral high inguinal orchidectomy (Intraoperative photograph)

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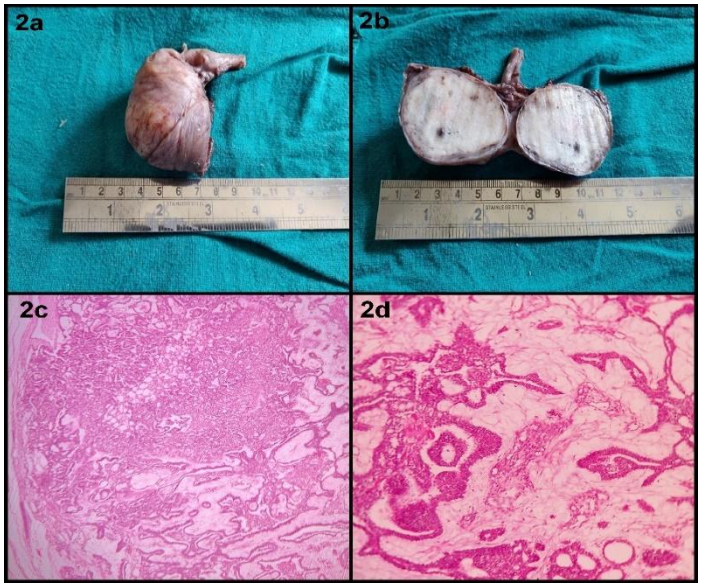

Fig. 2a & 2b: Grossly, the testis and spermatic cord measures 5.5x 4.5x 3.5cm and 4x1cm respectively. External surface is congested. The cut surface of testis showed a homogenous, grey white, circumscribed area measuring 4.5x 4.5cm. Few minutely cystic areas and pinhead size areas of haemorrhage are also seen. **Fig. 2c & 2d:** Microphotographs show a tumor composed of epithelial and mesenchymal elements arranged in microcystic, glandular- alveolar, perivascular and papillary formations. At places, solid cords of cells are also seen. The cystic spaces are lined by flattened, endothelium like layer of cells. Peivascular Schiller- Duval bodies are also noted. (H& E; 40x and 100x respectively)

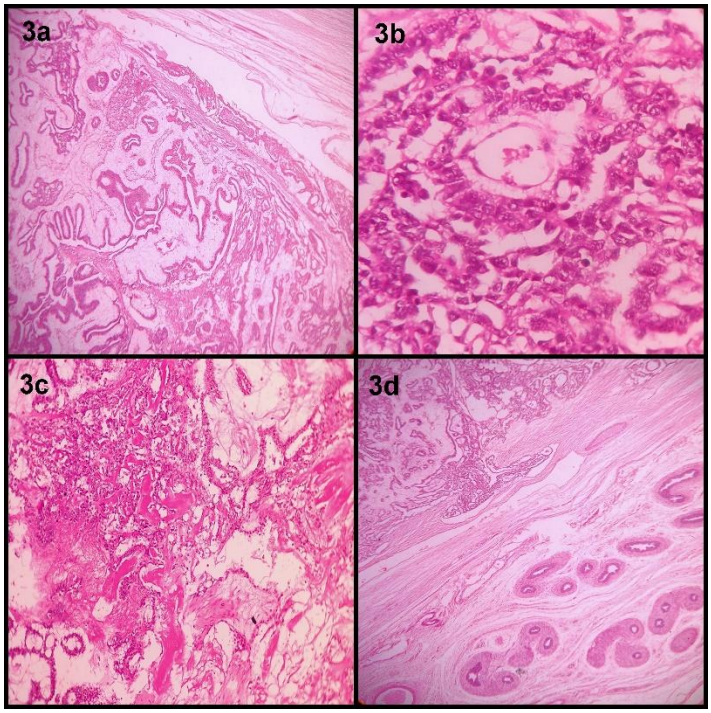

Fig. 3a: Microphotograph shows epithelial and mesenchymal elements arranged in reticular and microcystic pattern (H& E; 40x); **3b**: shows perivascular Schiller- Duval bodies which are glomerulus-like structures composed of a central blood vessel enveloped by tumor cells within a space that is also lined by tumor cells (H& E; 400x); **3c:** shows areas of intracytoplasmic and extra cytoplasmic eosinophilic inclusions along with occasional areas of haemorrhage and necrosis (H& E; 40x); **3d:** The tumor cells are seen infiltrating the tunica albuginea but not invading through it (H& E; 40x).

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