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RESEARCH ARTICLE

A CASE OF PARATESTICULAR TUMOR IN AN ADOLESCENT BOY

[1,*]Balasubramanian, A., [2]Bhanumati Giridharan, [3]Vasanth, M., [4]Shanmugasundaram, P.N. and [5]Sathishselvakumar

[1]MS, DNB, FMAS, Assistant professor, Department of General surgery, ESIC Medical College & PGIMSR, Chennai-78

[2]MS, Associate professor, Department of General surgery, ESIC Medical College & PGIMSR, Chennai-78

[3]Assistant professor, Department of General surgery, ESIC Medical College & PGIMSR, Chennai-78

[4]MS., Professor, Department of General surgery, ESIC Medical College & PGIMSR, Chennai-78

[5]MD., Assistant professor Department of Pathology, ESIC Medical College & PGIMSR, Chennai-78

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**Corresponding author:*
Balasubramanian, A.,

ABSTRACT

Primary paratesticular tumors are a rare group of tumors that present as a painless mass in the scrotum. Generally, due to the rarity and decreased awareness of their presence, they appear as an incidental finding on the operating table or in the postoperative pathology. Despite the fact that these tumors can be benign or malignant, they are entitled to be handled without preoperative biopsy. The presence of these tumors in younger individuals complicates management, and strong evidence is needed to justify orchidectomy in these cases. There has been a vast improvement in the literature regarding paratesticular tumors and their management strategies over the past few years which are a boon to the clinicians. We discuss here one such rare case of paratesticular tumor in an adolescent male and share our experience in its management.

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INTRODUCTION

Scrotal mass represents several inflammatory, benign and malignant pathologies. Pain is a useful symptom to delineate the inflammatory pathologies such as torsion testis and epididymoorchitis. While the painless scrotal masses claims the most representation in a general surgery outpatient services , the incidence of para or extra testicular masses is often under recognized and forgotten in the initial workup. The para testicular mass can arise from the tunica coverings, epididymis and spermatic cord. They contribute 7 - 10% of all intrascrotal tumors (1). Though 75% of these lesions are benign, the remaining 25% malignant pathologies pose great difficulty in identification and selecting the appropriate strategy of management (1). Here in this case report, we present a 17year old adolescent male with a painless scrotal mass.

Even with the identification of para testicular origin of the lesion, a multitude of benign entities in the differential diagnosis had left the possibility of preserving the testes alive. While benign lesions can simply be removed with testicular and fertility preservation, any margin of error could ruin the patient's chances of complete, one-time curable resection and land him with definite local recurrence. Due to the lack of uniform consensus in the treatment of paratesticular tumors, the management of this patient was exceedingly challenging. This article mainly highlights the approach and management of para testicular tumors and in particular about the rhabdomyosarcoma entity which is more common in this age group with its distinct pathological features

Case Presentation: A 17 year old adolescent male presented in our surgical clinic with complaints of swelling in right side of the scrotum for the past eight months. The swelling was painless and it had gradually increased in size over the past eight months. He denied any antecedent history of trauma. He

also did not have any prodromal symptoms of fever, sweating and weight loss. He was well built and adequately nourished for his age. The examination of scrotum revealed a 15 x 12 cm right sided scrotal mass extending into the root of the scrotum (Figure **1**). The mass had an irregular surface and borders, and the right side testis was not separately palpable from the mass. The mass had varied consistency with soft and hard areas. The fluctuation and trans illumination were absent. The examination of abdomen and left supraclavicular fossa were normal. With this history and clinical examination, our initial workup included an ultrasonogram and serum markers to rule out testicular malignancy and chronic epididymoorchitis. The high frequency ultrasonogram reported it as large right sided epididymal mass. The serum markers including beta-hCG, alpha fetoprotein and Lactate dehydrogenase (LDH) were within normal limits. As a result, the diagnosis was narrowed down to a paratesticular lesion. Further a magnetic resonance imaging (MRI) of the abdomen, pelvis and scrotum was done to identify nature of the lesion. The MRI reported a well-defined, encapsulated lesion of size 9.5 x 7.9 x 6.9 cm in the right scrotal sac with its epicentre in the distal spermatic cord (Figure **4**). The lesion appeared heterogeneous with solid and cystic components. There was no significant para-aortic and pelvic lympadenopathy. The report suggested a right sided distal spermatic cord tumor with a suspicion for neoplastic aetiology. Considering the possibility of benign lesions and the difficulty of obtaining a pre-operative biopsy to prevent scrotal violation, it was difficult to determine the nature of the lesion and explain the surgeon's decision to perform frozen control orchiectomy to the patient and parents.

After obtaining informed consent, a high inguinal approach was made and a noncrushing occluding clamp was applied over the proximal part of the cord near the deep ring. The intact encapsulated scrotal mass was delivered into the inguinal wound with gentle maneuvers involving traction (from above) and squeezing (from below). The mass was arising from the junction of the distal spermatic cord with epididymis and was composed of a hard fibrotic area surrounded with a jelly like myxoid tissues (Figure **2**}. An intraoperative frozen section was taken from both the areas, which was reported as highly suspicious for sarcoma. A high inguinal radical orchidectomy was done. The final postoperative histopathology report was suggestive of embryonal rhadomyosarcoma with positive desmin, vimentin, myogenin& CD56 on immunohistochemistry (Figure **3**). Subsequently a whole body PET-CT (Positron emission tomography with computed tomography) was done and reported no evidence of active disease (Figure 5). Considering the disease pathology of rhabdomyosarcoma, an adjuvant chemotherapy with vincristine, Adriamycin and Cyclophosphamide was given. The patient had completed the adjuvant therapy and is on regular follow-up without any recurrence.

DISCUSSION

The term Para testis encompasses the testicular tunics, epididymis, rete testis, spermatic cord and its related embryological remnants in the scrotum. The Para testicular masses are mostly neoplastic.

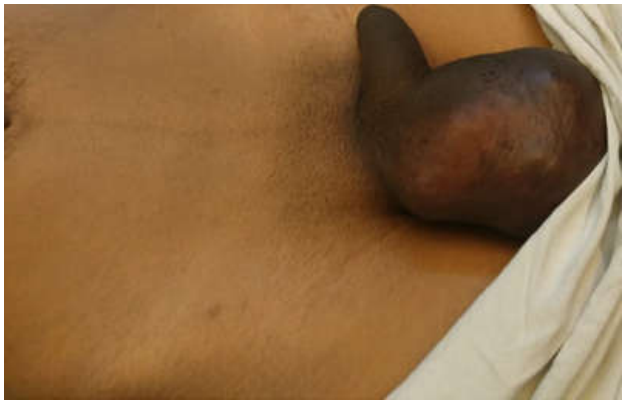

Figure 1. Preoperative picture showing right side scrotal mass

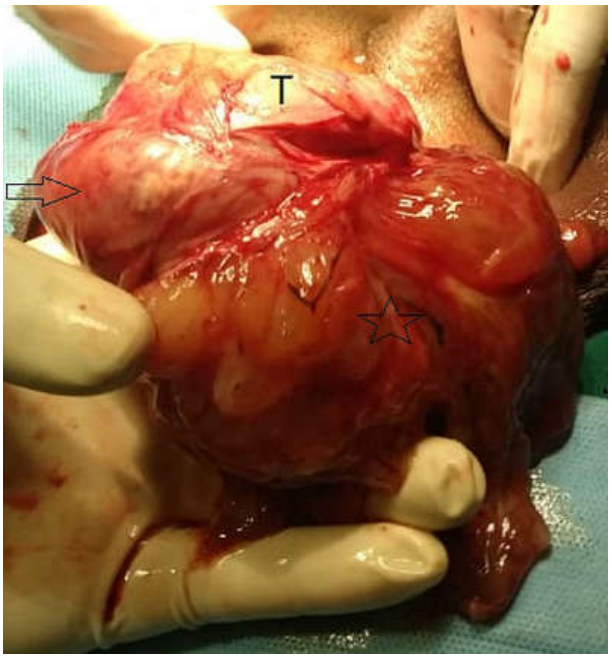

Figure 2. Intraoperative picture showing the testis (T) and the Paratesticular tumor arising from the inferior part of spermatic cord with hard (arrow) and myxoid (star) areas

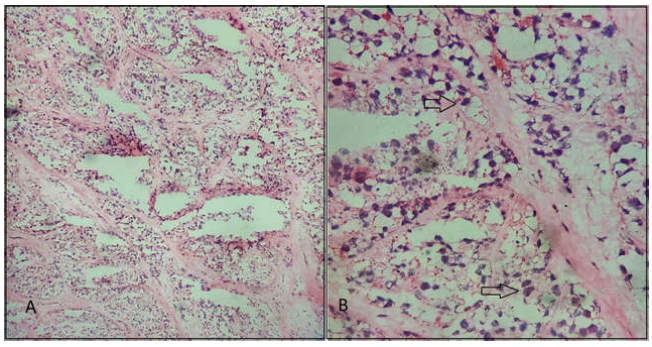

Figure 3: Histopathology of the tumor with low power (A) and high power (B) views showing the nests of malignant cells (Arrows)

The non-neoplastic Para testicular lesions (6- 30 %) include congenital cysts, vascular lesions, inflammatory lesions, ectopic tissues and fibrous pseudo tumors (2). The neoplastic lesions take origin from mesenchymal, epithelial and mesothelial cells depending on the site of origin. The spermatic cord is the most common site of tumor origin accounting for more than 75% of all paratesticulartumors.

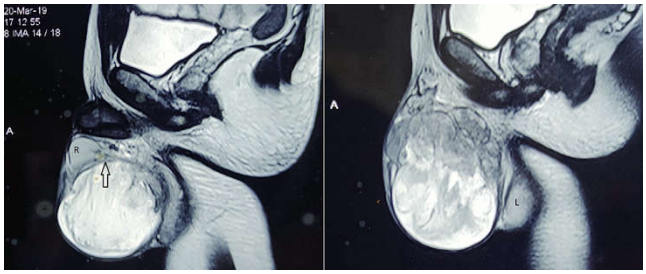

Figure 4. Preoperative MRI showing Left testis (L), Right Testis (R), and the right paratesticular lesion arising from the inferior end of spermatic cord (arrow). The lesion appear as well defined encapsulated, heterogenous mass with solid and cystic components

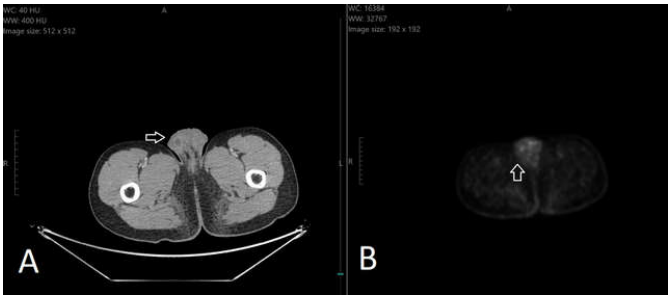

Figure 5. Post operative whole body PET CT showing the post operative inflammatory changes(A) in CT and the associated metabolic uptake (B) in PET

Among the neoplastic lesions most are benign with lipomas, adenomatoidtumors and leiomyomas being common. However about one third of the neoplastic tumors are malignant and 90% of them are sarcomas. The liposarcoma, leiomyosarcoma and rhabdomyosarcoma are the most common variants in the sarcomas occurring in this region (3). Spermatic cord tumors are of prime importance because they contribute the most of the para testicular tumors. They mostly arise from the inferior part of the spermatic cord, making them appear as an intrascrotal mass (1). They present usually as a slow growing solid intrascrotal mass. The presentation is more of a nonspecific pattern that it can mistaken for a chronic epididymo-orchitis, tuberculosis or for a testicular tumor. The diagnosis with clinical examination alone can miss many of them. So an inclusion of high frequency ultrasonography (USG) is a must to discern the paratesticular origin of the tumor. Added to it we may need an MRI (with contrast) of the scrotum to have better characterization of the tumor. Any suspicion for malignancy requires screening of both the regional lymphnodes (retroperitoneal and iliac nodes) and the chest as a part of metastatic workup. Serum markers related to testicular malignancies are also usually done to rule out testicular malignancies.

The most common malignant pathology is liposarcoma followed by leiomyosarcoma and they usually present as a well differentiated and slow growing tumor in the fifth or sixth decade of life. The paratesticular rhabdomyosarcomas (PT-RMS) are very rare tumors but their incidence is more in the first two decades of life. Considering the site of involvement, the paratesticular rhabdomyosarcoma is the third most common in the genitourinary system following the prostate and urinary bladder (4). Conventionally three histological types of rhabdomyosarcoma has been described namely pleomorphic, alveolar and embryonal, the latter being more common and has the best prognosis.

The rhabdomyosarcomas have the highest propensity for lymph nodal and haematogenous spread than any other sarcomas. The age distribution is bimodal with peaks at 1-5 years and 16 years of age. The presentation is nonspecific with a painless slow growing scrotal mass. The imaging features with Ultrasonography and MRI are also inconsistent and nonspecific, though they commonly appear as a heterogeneous mass with variable echogenicity (5). Histopathology along with Immunohistochemical markers (Myogenin & Desmin) for skeletal muscles forms the basis of diagnosis. However the luxury of preoperative biopsy is unavailable for paratesticular tumors in view of scrotal violation. The younger age of presentation with the limited information from the imaging modalities make the straight forward decision of radical orchiectomy questionable and unacceptable. The need for confirming the diagnosis intraoperatively with a frozen section is a must and answers all the debatable questions around it (6). Also equal importance must be given for not violating the scrotal planes and the covering of the tumor , which may disseminate the tumor.The presentation in our case was very much similar and the thoughtful inclusion of preoperative MRI and intraoperative frozen section balanced our approach for a paratesticulartumor wherein both benign and malignant lesions can mimic each other. The ultimate treatment goals in case of a malignancy are local and regional control. While a high inguinal radical orchiectomy takes care of the local control, the strategy for regional control is still full of controversies. The role of ipsilateral retroperitoneal lymph node (RPLND) dissection is much very debated and at present is advocated only for those patients having CT detected lymphadenopathy as per the fourth Intergroup rhabdomyosarcoma study (IRS-IV) (4). However in a subset of adolescent males with age more than 10 years and tumor size more than 7cm the likelihood to have retroperitoneal lymph node spread is high and so ipsilateral RPLND is strongly recommended in them. The fourth intergroup rhabdomyosarcoma study (IRS-IV) provides the risk categorization of Rhabdomyosarcoma by using the TNM-8 classification, post-operative resection status and histopathology types (4). Also the role of whole body PET CT has been recently well documented for staging, restaging and response monitoring in pre and post-operative settings with an accuracy of 86% (4). Chemotherapy works best for rhabdomyosarcoma in all stages and vincristine, dactinomycin, and cyclophosphamide are used with good results as a systemic polychemotherapy. Hence adjuvant chemotherapy can be considered in all stages of rhabdomyosarcoma (1). The role of radiotherapy in paratesticular sarcomas is currently reserved only for high grade tumors and as a salvage treatment for nodal disease with no recommendation in localized diseases. The follow-up requires minimum of 36 months because of the high rates of local recurrence (1).

CONCLUSION

An estimated 30 percent of paratesticulartumors are malignant (mostly sarcomas), making preoperative imaging, including a high-frequency ultrasound and MRI, as well as whole body PETCT, essential. There is a high incidence of rhabdomyosarcomas among adolescents and these tumors pose a diagnosis problem because of their nonspecific clinical and radiological findings. Considering the higher chance of benign lesions, frozen sections should be considered in intraoperative diagnosis before orchidectomy. Malignant tumors diagnosed by frozen biopsy are removed surgically by the high inguinal approach with orchidectomy, while regional nodal clearance

depends on specific risk factors. Furthermore, rhabdomyosarcomas usually respond better to chemotherapy, making adjuvant therapy an option at every stage of the disease. In the end, a comprehensive management plan for each patient will depend on greater awareness and understanding of paratesticulartumors and consensus on their management. Such an individualized plan of management will lead to better results and greater patient compliance.

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