CASE REPORT

Burned-out Metastatic Yolk Sac Tumour: A Rare Case Report

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Abstract

Burned-out testicular tumor is a very rare clinical entity. There are no clinical findings in the testicle because it regresses spontaneously without any treatment and generally presents with metastases. Burnt out testicle is often seen with Yolk sac tumour. The diagnosis of yolk sac tumours is usually a multimodality diagnosis including a biopsy of the lesion. We describe a case of metastatic yolk sac tumours that presented with deposits in inguinal lymph nodes. No testicular tumour was identified in our case.

Keywords:

Testicular tumor, Yolk sac, Metastatic

Introduction

Testicular tumors are uncommon in children, comprising approximately 1% to 2% of all pediatric malignancies.[1] Yolk sac tumors are the most common among testicular tumors.[2] The prognosis of testicular yolk sac tumors is dependent on early detection and treatment. Pediatric patients with testicular yolk sac tumors usually present with an asymptomatic scrotal mass early (Stage I) in the disease process.[3] Evaluation of the solid scrotal mass includes: scrotal ultrasound; chest, abdominal, and pelvic computed tomography (CT); and determination of serum tumor marker levels such as alpha-fetoprotein (AFP) and beta-human chorionic gonadotropin (b-hCG) and histopathological evaluation. An elevated serum AFP level is closely associated with yolk sac tumors in more than 90% of patients.[4] In this article, we present a rare caseof with yolk sac tumorin 13 year old young boy who presented with deposits in the inguinal and axillary lymph nodes. No testicular mass was identified in our case.

Case presentation

A 13-year-oldboy was admitted to pediatrics ward with complain of abdominal distension and generalized lymphadenopathy since few months.On examination inguinal and axillary lymphadenopathy was noted. Ultrasonography guided FNAC of inguinal and axillary lymph nodes was then performed. Cytological examination from both the sites showed similar morphology. FNAC from bilateral axillary and inguinal lymph nodes findings: Microscopy: Smears were highly cellular showing malignant cells in clusters and sheets. (*Fig 1A*) Few cells were lying individually scattered. These individual cells showed moderate amount of cytoplasm with focal vacuolization, central nuclei with frequent binucleation to multinucleation, vesicular chromatin, conspicuous nucleoli and moderate degree of pleomorphism. Background appeared to be mucoid to granular and hemorrhagic along with scattered mature lymphocytes. (*Fig 1B*) The diagnosis of metastatic deposits of malignant tumor in inguinal and axillary lymph nodes was made. Possibility of Germ Cell Tumor was suggested.Subsequently, an excised axillarylymph node was received in our histopathologysection.

Excisional biopsy findings:

Gross: Received single globular soft tissue piece measuring 3 x 2.5 x 1 cm. External Surface was smooth

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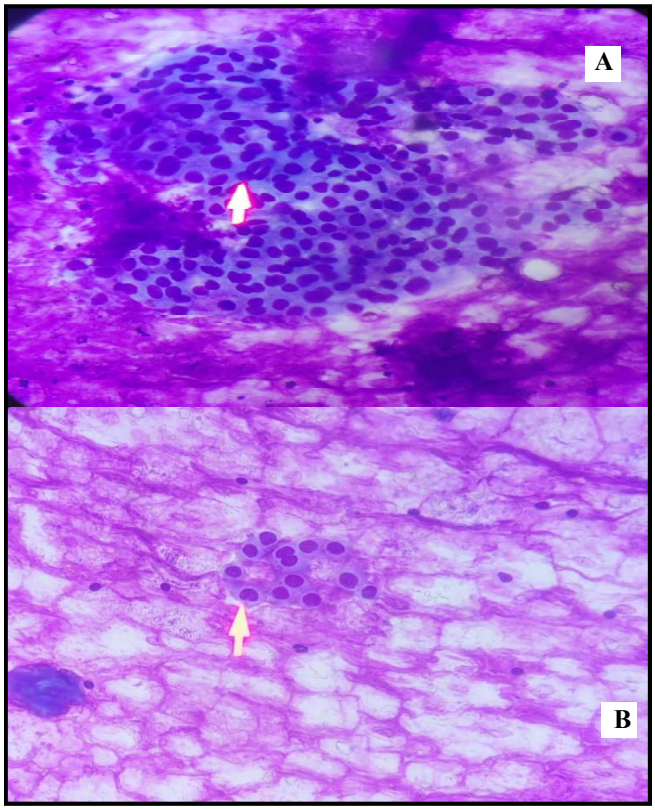

Fig No.1 (A) Cellular showing clusters of malignant cells. The individual cell showing moderate amount of cytoplasm with focal vacuolization, central nuclei. Many cells showed binucleation to multinucleation (arrow), vesicular chromatin, conspicuous nucleoli and moderate degree of pleomorphism. (B) A small cluster of malignant cells showing central round nuclei with cytoplasmic vacuolization (arrow). Background is mucoid to granular with haemmorraghic background with scattered mature lymphocytes.

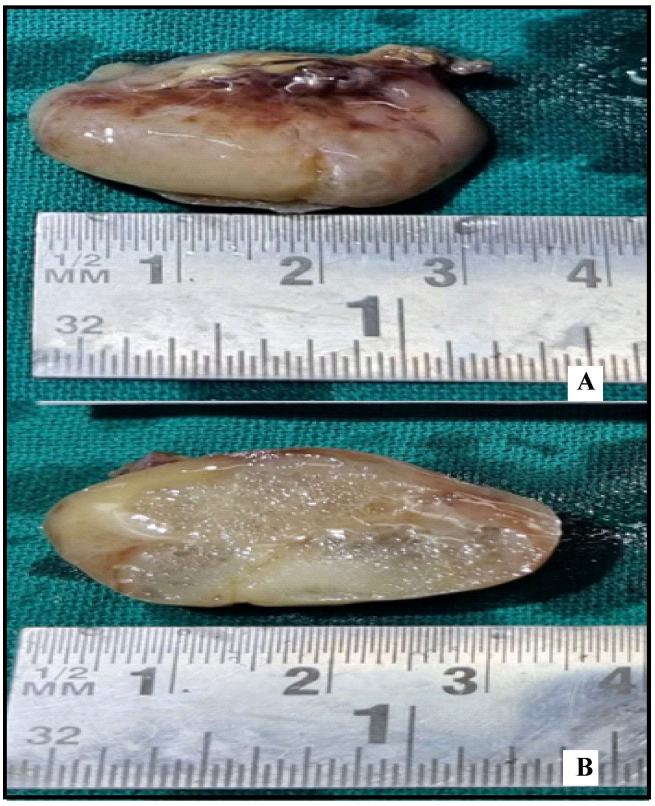

Fig No. 2 (A) Excised axillary lymph node measuring 3 x 2.5 x 1 cm. with outer smooth surface (B) Cut section showing homogenous white yellow gelatinous area

and On cut section, homogenous white yellow gelatinous areas were identified (*Fig 2A, B*). **Microscopy:** Sections revealed almost completely effaced lymph node by infiltrating tumor cells forming glands and vesicles lined by flat to columnar epithelial cells with moderate pleomorphism (*Fig 3A, B*). At places these vesicles coalesced to form macrocysts. Epithelial papillae and hobnailing of nuclei was present. In view of morphological features, pediatric age group and generalized lymphadenopathy the diagnosis of Metastatic Testicular Tumor (Yolk Sac Tumor) was kept. Testicular imaging studies did not reveal any testicular mass but was completely fibrosed. Serum alpha fetoprotein was elevated to 38 ng/ml (normal range 10-20 ng/ml).

Discussion

Pre-pubertal testicular tumors represent 1-2% of all solid pediatric lesions with an incidence of 0.5-2 per 100,000 children. Paratesticular tumors account for only 15% and are mostly benign.[4] Malignant paratesticular tumors do arise, the most common being rhabdomyosarcoma, a non-germ cell tumor (GCT). Ultrasound (US) and tumor markers aid in the diagnosis and treatment planning. Correct diagnosis is usually made only from histopathological examination of the excised specimen. Yolk Sac tumor is the most common malignant histology in children and is common at sacrococcygeal, retroperitoneal, mediastinal and prepubertal testis location. Different histologic patterns are common in a single tumor and 25% of childhood tumors contain more than one histology. However burnt out testicular tumors are rarely reported. Burned-out tumour of the testis is a very rare clinical entity. The term 'burned-out' tumour of the testis describes a spontaneously and completely regressed testicular tumour with no treatment. It presents by metastases to the retroperitoneum, mediastinum, lymph nodes, lungs and liver.[5] Ultrasonographic findings in burned-out tumours are raised echogenicity in a focal area, probably due to calcium deposits and fibrosis. There

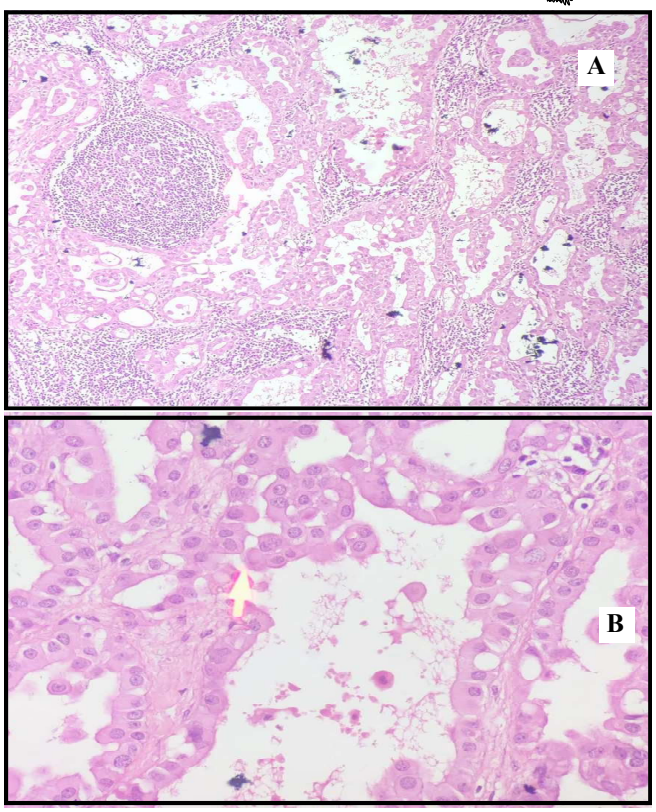

Figure No. 3 (A) Section showing polygonal tumor cells surrounding a small blood vessel (100X; H&E). (B) The individual tumor cells show moderate amount of cytoplasm with focal vacuolization, central nuclei with frequent binucleation to multinucleation, vesicular chromatin, conspicuous nucleoli and moderate degree of pleomorphism (400X magnification).

are no tumoural findings on pathological examination of the testis. Despite ultrasonographic findings being normal, testis biopsies should be performed if there is any suspicion, and/or risk factors for in situ malignancies. Retroperitoneal germ cell tumours appear to arise from primary testicular lesions. Primary extragonadal germ cell tumours of the retroperitoneum are probably a rare entity. They should be considered to be metastases of a viable or burned-out testicular cancer until proven otherwise histologically. Fabre *et al* declared that there is variable clinical presentations of patients with "burned-out" testicular tumours, and that this diagnosis, although infrequent, must be considered and extragonadal germ cell tumours should be considered to be metastases of a "burned out" testicular tumour, and must be investigated.[6] In the presence of retroperitoneal lymph nodes, a testicular ultrasound examination can detect tiny intratesticular lesions, minimizing the possibility of a primary extragonadal germ cell tumour. It is important to distinguish 'burned out' tumours of the testis from true extragonadal germ cell tumours because primary removal of the testicular tumour is necessary for treatment.[7] Orchiectomy is generally completed with cisplatin-based combination chemotherapy protocols. This therapy is very effective in the treatment of seminomas and non-seminomatous germ cell tumours. Combination chemotherapy with bleomycin, etoposide and cisplatin is also effective and no disease recurrence is observed.

To conclude, Scrotal sonography is very important for the detection of intratesticular lesions, especially in patients with extragonadal metastatic involvement and normal palpation findings for the testis. A burned-out testicular tumour should be considered when punctuate echogenic foci are seen without any evidence of hypoechoic mass lesions. Testicular biopsy should be performed if there is any risk factor for malignancy.

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Conflicts of Interest

There are no conflicts of interest.

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