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Case Report

Testicular Fibro- Sarcoma in Pediatric Patient

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Abstract

Sarcoma of the testis are extremely rare tumors, their incidence being difficult to assess accurately. a case of two-year-old male, presented with painless scrotal swelling that increase in size insidiously within three months, send to urology, the examination revealed left testicular swelling –hard, not tender and oval shape. scrotal US, show large mildly hyperechoic mass fat echogenicity seen in the left inguinal canal and scrotum measuring about 84x41x44mm diameters. CT chest and abdomen were normal. Radical orchidectomy was done through an inguinal approach, histopathology showed morphology constant with fibrosarcoma of gonadal stromal origin. After 10 months, noticed reappearance of swelling in the left hemiscrotum.

CT scan abdomen and pelvic showed moderate to gross ascites with cystic lesion in the pelvis on the left side, these findings are highly suggestion a of malignant ascites with possibility of metastatic cystic lesion. excision was done, Histopathology revealed spindle cell sarcoma morphology favor fibrosarcoma of gonadal stromal origin. chemotherapy start for this case with ifosfamide+ doxorubcin protocol one cycle, his condition progress with hug ascites patient died. In conclusion, the recurrence rate of testicular sarcoma is high following radical orchidectomy, prognosis is very poor.

Keywords: Testicular tumor, testicular fibrosarcoma, spindle cells, recurrent testicular sarcoma, prognosis

Introduction

Testicular cancer represents about 1.5% of male neoplasms and 5% genitourinary tumor. Most testicular tumor are diagnosed in 20th and 30th years of life. the histological type varies, although there is clear predominance (90-95%) of germ cell tumor. [1] infantile fibrosarcoma is rare in early childhood malignancy .it includes proximality 10% of all sarcoma in children [2]. Very little epidemiological data regarding soft tissue sarcoma and bone sarcomas from Yemen in general and Aden region in particular are available, one cross sectional study was carried out at Nation Oncology Center, Aden Yemen from 2009-2013 out of 107 new diagnosed patient 54 belonged to bone sarcoma and 53 to soft tissue sarcoma, the most common site of origin was lower extremities 55.1%. retroperitoneal 11.2%, trunk 10.3% and upper extremities 9.3% [3]. Furthermore, the rarity of primary fibrosarcoma in the further justifies the following report.

Case Report

a case of two-year-old male, presented with painless scrotal swelling that increase in size insidiously within three months, send to urology, the examination revealed left testicular swelling - hard, not tender and oval shape. Scrotal US show large mildly hyperechoic mass fat echogenicity seen in the left inguinal canal and scrotum measuring about 84x41x44mm diameters. CT chest and abdomen were normal. Radical orchidectomy was done through an inguinal approach ,12/1/2018 histopathology showed morphology constant with fibrosarcoma of gonadal stromal origin. Panel of immunostains is needed: vimentin, pankeratin, EMA, inhibin, CD99, S100, SMA. Desmin. but family patient neglected to do it. after 10 months, noticed reappearance of swelling in the left hemiscrotum.

Excision was done

11/11/2018 Histopathology revealed spindle cell sarcoma morphology favor fibrosarcoma of gonadal stromal origin Immunotoxins were done: the tumor cells are positive for CD 99 and negative for pankarrtin, vimentin, SAMA, S100, CD34 and BCL2.

Ct scan chest

12/11/2018:no significant pleuropulmonary or mediastinal abnormality CT scan abdomen and pelvic 12/11/2018: showed

moderate to gross ascites with cystic lesion in the pelvis on the left side, these findings are highly suggestion a of malignant ascites with possibility of metastatic cystic lesion (Figure 1). Chemotherapy start for the case with Ifosfamide +doxorubicin protocol one cycle, his condition progressive with hug ascites patient died.

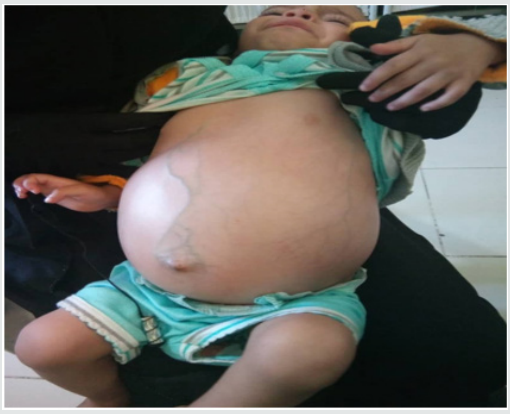

Figure 1: case of testicular fibrosarcoma.

Discussion

Sarcoma of testis are extremely rare, when you review the literature of published series of sarcoma they were distributed as fellows: leiomyosarcoma 32%. Rhabdomyosarcoma RMS,24% and liposarcoma 20% are the comments subtypes [4,5]. Gowing and Morgan studied all mesenchymal tumors arising in testicular tissues, and their relative rarity is apparent; the study consists of 22 sarcomas enrolled in a survey of around 1000 patients with testicular neoplasm. The majority of the sarcomas in this study were mesenchymal, that is fibrosarcoma, liposarcoma, myosarcoma, rhabdomyosarcoma and leiomyosarcoma, or combinations [6]. The sarcoma may present with a unilateral, painless scrotal swelling with short duration as seen in this case report

The commonest way of metastasis is via lymphatic to internal iliac and para -aortic lymph nodes. Distance metastasis to liver, lungs and bone by hematogenous spread accounts for 20% of patients in initial presentation [7]. Management of testicular fibrosarcoma has shifted to multispecialty approach that improves the survival chances up to80% by incorporating chemotherapeutic medication, radiotherapy or surgical intervention [7,8]

In this case reported the radiological staging was N0M0, so preferred fellow up after radical orchidectomy. inguinal approach orchidectomy with ligation of spermatic cord at the internal ring is the standard surgical intervention as seen in the case. The site of tumor origin (, testicular-Para testicular), lymph node involvement, distant metastasis, presence or absence of residual disease after surgical resection will help in staging of testicular and the plan of management of fibrosarcoma of testis. Gilbert, quoted by O'Brien (1942), accepted seven cases of primary fibrosarcoma out of 91 malignant epididymal neoplasms in the literature. However, we do not know the cases which he accepted or the criteria upon which he judged them [9].

Catton et al. [10] mention 14% local recurrence in patients who underwent wide local excision and /or radiotherapy after original excision of testicular or paratesticular sarcomas. They also suggested that orchidectomy alone is inadequate for testicular sarcomas and recommended adjuvant wide hemiscrotum excision, which may include inguinal lymph nodes for those managed with orchidectomy or local excision. they found that there was microscopic disease present in 27%, following wide excision in patients with apparently completely excised tumors. In current case, after 10 months there was noticed reappearance of swelling in the left hemiscrotum, which made us to think of local recurrence, and therefore the decision of re-exploration was made with wide local excision of the swelling and the hemiscrotum. Histopathology revealed spindle cell sarcoma morphology favor fibrosarcoma of gonadal stromal origin Immunostains were done: the tumor cells are positive for CD 99 and negative for pankarrtin, vimentin, SAMA, S100, CD34 and BCL2. CT scan abdomen and pelvic showed moderate to gross ascites with cystic lesion in the pelvis on the left side, these findings are highly suggestion a of malignant ascites with possibility of metastatic cystic chemotherapy start for the case with Ifosfamide +doxorubcin protocol one cycle, his condition progressive with hug ascites patient died.

The role of retroperitoneal lymph node dissection (RPLND) in testicular sarcomas remain controversial [11-13]. Catton et al [11]. Recommended RPLND in their earlies series of 21 patients, but in their subsequent reported they noted that the patients with high risk of nodal metastasis are also that at high risk of concurrent or subsequent systemic disease; accordingly, they felt that those patients would benefit from systemic rather than regional treatment [10]. Other suggest that RPLND should be limited to patients with suspicion of LN metastasis in the image [14]. Adjuvant chemotherapy in testicular sarcomas is not fully established. in pediatrics, with testicular sarcomas -especially rhabdomyosarcoma (RMS)- adjuvant chemotherapy had good outcome on the overall survival [15,16]. It uses in adult testicular sarcomas has not been yet specially addressed in controlled study [17]. The use of post -intervention chemotherapy for adult -grad testicular fibrosarcoma is controversial meta-analysis of randomize trials of testicular sarcomas at different centers showed that doxorubicin-based adjuvant chemotherapy significantly improved the time to local and distant failure [18].

Conclusion

Testicular fibrosarcoma is extremely rare testicular tumor, usually indolent course with potential for distant metastases. The diagnosis of pure fibrosarcoma should be made only after extensive sampling of the testicular tumor to rule out an associated germ cell component. the differential diagnosis also includes the fibroma of gonadal stroma origin and the unclassified sex cord -stromal tumor with predominance of spindle cells

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