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Paratesticular Liposarcoma: A Case Report and Review of Literature

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Abstract

Paratesticular liposarcoma, a rare pathological entity. They usually mimic the inguinal hernia because of the location of the tumor. Physical examination is not considered appropriate for diagnosis; hence, for confirmatory diagnosis ultrasound and CT scan is performed. A 66 year old heavy smoker came into the outpatient department with a painless swelling on left side. Initially no suspected metastatic lesion; however, detailed investigation revealed small hepatic and renal cysts with lymphadenopathy. The dedifferentiated histopathological report is suggestive that it is high grade tumor; hence, patient underwent radical orchiectomy with the excision of the paratesticular mass followed by chemotherapy and lymph node dissection.

Keywords

Paratesticular liposarcoma; Tumor; Chemotherapy; Lymph node; Ultrasonography; Lymphadenopathy

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Introduction

Scrotal mass is an outgrowth extension observed in males, which is a challenging task for the doctors to identify and remove. The scrotum is a male reproductive organ enclosing numerous structures including germinal centres, epididymis and spermatic cord. Any mutation in the epithelial cells usually produces an abnormal mass of cell infiltrating soft tissue. The scrotal mass is divided into two types, testicular and paratesticular. Testicular masses are identified as lump of tissues in the testis while the paratesticular masses are characterized as slow growing, size ranging from 0.5-5 cm benign tumor with 30% adenomatoid type while 77% of benign tumors are located in the epididymis [1]. This type of tumor is generally observed in age 20-40 years [2]. The benign tumors always exhibit a chance for malignant transformation; in the case of liposarcoma the malignant transformation is rare in adults. These include adenocarcinomas, rhabdomyosarcoma, leiomyosarcomas, and liposarcomas.

Liposarcoma, is second most common soft tissue tumor in adults arising from the primitive mesenchymal cells in the adipose tissue. Occurrence of testicular liposarcoma only accounts for 12% of all liposarcoma while the extension of this tumour from a spermatic cord is only observed in 3-7% of cases [3]. Most cases reported the slow growing tumour mass towards the inguinal area as a common pathway to spread locally commonly at the age of 55 years [4]. To identify correct location of the mass is a critical factor that is achieved through history and proper examination. Series of radiological examination is required for differentiating a mass, which includes ultrasonography for mass location; doppler feature to evaluate blood flow in the mass and CT/MRI scan [5]. Clinically a differential diagnosis through imaging is a valuable way to differentiate the tumours. Ultrasonography shows a heterogeneous echogenicity of spermatid cord liposarcoma but no specificity defined. CT scan at this point is necessary for a definitive diagnosis in nearly half of the cases that determine the involvement of the anterior abdominal wall and/or retroperitoneum. A similar case was reported in present study report which described a case of liposarcoma in the spermatic cord with a detail literature review of spermatic cord liposarcoma.

Case Report

Sixty six years of Libyan patient presented with a left painless scrotal swelling for the past 4 months in the outpatient department. The swelling was clinically examined and an ultrasound was performed to categorise the tumour mass. In physical examination, the swelling was large, firm and was interpreted as a testicular tumor. The ultrasonography revealed left enlarged testis with well-defined paratesticular heterogeneous mass. The patient was planned for the left orchiectomy along with excision of Para testicular mass.

The surgeons proceeded with the dissection of the spermatic cord. Intraoperatively, a multi-lobulated, encapsulated, round mass was isolated carefully for excision; however, the mass was firmly attached to the surrounding soft tissue structures. In certain cases the mass moves towards the inguinal area, portraying as inguinal hernia, which can lead to misdiagnosis. Nevertheless, the mass was distinguishable in this patient; yellowish lipoma like texture, size 10×8×7 cm firm and palpable. The treatment proceeded with high inguinal left orchiectomy in addition to the excision of paratesticular

mass. The mass was send for the pathological analysis. The histological examination revealed de-differentiation paratesticular liposarcoma with evidence of myxoid degeneration, with mitotic activity (0-3 mitoses/10 HPF). The immunohistology demonstrated a raised level of S100 protein with lipomatous nature while negative staining for CK. The past history did not present with any doubt related to metastatic lesion at the time of admission; however, to further confirm any metastatic lesion, the patient underwent MRI, which exhibited multiple small hepatic and renal cyst formation. It was reported that 3-4 cm size masses were seeded in the lung along with lymphadenopathy in the mediastinum. The post-operative period was uncomplicated stage and patient was discharged on the 2nd postoperative day.

Discussion

The paratesticular liposarcoma is a rare tumour arising from primitive mesenchymal cells in the male reproductive part. The scrotum is a genital part containing epididymis and spermatic cord that extends to form benign tumours in rare conditions accounting for 3-7% cases [6]. These paratesticular tumours are inseparable from testicular type. Vinayagam et al. [5] pointed out that total 186 cases have been identified until now; nevertheless, further investigation is necessary. In the majority of cases, it is difficult to determine the location; however, 90 % of origin arises from spermatic cord [6].

Liposarcomais benign tumors arising from the adipose tissue anywhere in peritoneum and abdomen. Only 12% arise from paratesticular tissue with 70 % cases to be benign [7]. Cases have presented with well differentiated slow growing painless masses towards the inguinal area. Some studies displayed manifestation mostly in the age group fifth to the sixth decade, in contrast, others tumour presenting in the age group 20 -40 years [3,8].

Liposarcoma of the spermatic cord is commonly observed as a slow growing lipoma like texture painless scrotal swelling. Authors suggested that the right preponderance at an average age of 55 is a common presentation in many patients [9]. Few patients (6%) have reported a history of scrotal surgery and trauma that was considered as a triggering factor for seeding of mutated cells (3). As the swelling travels towards the upper side, it often causes misdiagnosis, with assumption of inguinal hernia, hydrocele, spermatocele, or a tumor of the testis and epididymis. These types of tumors are rarely diagnosed through physical examination. Proper imaging is necessary to identify the location and proceed with a definite diagnosis such as CT scan, MRI and ultrasonography.70% of tumor is benign while 30 % of malignant cases are often known as sarcomas [9]. The first priority suggested by many surgeons is scrotal ultrasound and measurement of serum tumor markers including beta-human chorionic gonadotropin, alpha-fetoprotein, and lactate dehydrogenase. Furthermore, the sonographic image presented that the mass in our patient was in hemiscrotum area, the fatty mass. It was suggestive that it is a liposarcoma. Therefore, confirmatory diagnosis was achieved through the CT scan and MRI. The patient's reports displayed a non-homogenous mass of fat with variable echoes. Therefore, literature suggests that the CT scan with contrast IV is considered the best radiological tool for diagnosis both preoperatively for staging and postoperatively during follow-ups [10].

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After the excision of the mass histological examination is the utmost in each case for grading and staging of the tumor. The present case histological report indicated a para testicular tumor mass in testis. The paratesticular tumor has been classified into three categories: well differentiated, pleomorphic and myxoid (round cell) type [7]. The well differentiated type often grows slowly in adipose tissue mostly in the retroperitoneum area. Histologically they display inflammatory and spindle cells with sclerosing activity. De-differentiated type share resemblance with well differentiated type and high grade non lipogenic tumour [11]. Microscopically these types have a fibrillary component in growing with numerous collagen fibres. Special stains do not exhibit an essential role; except, S100 protein stain positive for the lipoblast [12]. In pleomorphic type liposarcoma the mass contains a heterogeneous hyper and hypoechoic nodule that is distinguishable from the lipoma [10,13]. Nevertheless, well differentiated mass comprises of fat enclosed by thick septa with a similar appearance to lipoma. Generally, liposarcomas are classified based on histological appearance liposarcomas; myxoid accounts for 40% (most common), round cell, well differentiated (subdivided into lipoma-like, sclerosing, inflammatory and dedifferentiated), and pleomorphic [10,14]. Liposarcoma has close resemblance with lipoma especially with the lipoblastic cell types; however, their surface displays a mucinous appearance. Considering the large masses, they exhibit multinodularity, and multilocularity of fats and cartilage. Mostly, the 48.7% accounted masses are low grade well differentiated followed by 25.6 % myxoidliposarcoma [6]. Dedifferentiated type and pleomorphic type are known to be malignant in nature incidence accounting for 17.9% and 5.1%, respectively. Few authors were able to report unusual cases such as mixed type liposarcoma with well differentiated major pleomorphic and minor sclerosing components and welldifferentiated myxoid with angiolipoma [4,12]. The immunohistochemical markers are applied in each case for morphological classification.

A presurgical treatment commonly known as the needle biopsy is performed to diagnosis and classifies the tumor according to the stage and grade [15,16]. The lipomas are excised through the basic procedure; however, in the case of liposarcoma radical orchiectomy is performed. In the present case, the paratesticular liposarcoma is treated by radical orchiectomy with high ligation of the spermatic cord. Currently, efforts are being made to conduct partial orchiectomy to preserve testis. Radical orchiectomy involves removal of testis and spermatic cord for complete lysis of the tumour. According to Mullinax et al. [17], the randomised trial displayed 4 patients out of 38 underwent tumour resection, 2 patients with retroperitoneum involvement underwent tumor resection in addition to radical orchiectomy. In all cases, the tumor spread through direct invasion with local recurrence. Nevertheless, one patient presented with the obstructed left colon and nonfunctioning left kidney function underwent multiple organ re-section along with excision of liposarcoma [17]. Certain complications are associates with this procedure such as the hematoma formation, Ilioinguinal nerve injury during the nerve separation from the spermatic cord and inguinal hernia. It is always considered for the patient to undergo lymph node dissection in a case where the lesion has metastasized; however, it is avoided in the absence of lymphadenopathy despite a 15-20% chance of occurrence [16].

Previous cases have demonstrated that well differentiated masses have a capacity to recur even after complete removal. Certain pathological features are necessary to be noted which triggers the chance of recurrences including large tumor size, inguinal location, degree of nuclear differentiation, and depth of

invasion. 90% of the cases reported high recurrence depending upon the size, subtype and margin state [16]. Any lesion less than 1 cm with a negative margin indicates recurrence free removal [18]. Literature suggests any likelihood of positive surgical margins increases the risk for local recurrences and metastatic lesions. In certain studies the use of polychemotherapy (Vincristine, Cyclophosphamide and Doxorubicin) is advocated in high grade metastasis to reduce the local recurrence after orchidectomy that accounts for 25% to 37% [3,5]. The current case exhibited a high grade de-differentiated histopathology that is considered as poor prognosis in terms of survival and recurrences. Depending upon the size margin grade and subtype the survival rate and prognosis can be predicted; hence, only 20-80% demonstrates 5 year survival. Recurrence has been reported in 46-57% cases of paratesticular masses. Myxoid and well-differentiated demonstrates better prognosis than round cell and pleomorphic liposarcoma in many reported cases [6,19]. Five-year survival rates are 80% and 20%, respectively suggestive by previous case reports [6,14].

There has always been a controversial debate over the adjuvant therapies necessary after surgical removal. Numerous studies presented that combine therapies have always shown beneficial results. Liposarcoma is categorized as a radiosensitive tumor; therefore, surgical treatment followed by radiotherapy reduces the chances of relapse [3]. Regarding the radiation dosage, there is paucity of data. However, only selected mentions are recommended for radiotherapy; those who have high grade tumor and recurrence [20]. In the present study, patient was not recommended radiotherapy because of the tumor was completely excised. There is no insight for chemotherapy initially but after the reports were released the distant metastasis encouraged for some sessions with periodic follow ups are advised. The studies have displayed a recurrence rate at a period of 3.3 years with a range of 1 to 6 years [5]. Literature has shown in several metastatic cases that radiotherapy do not exhibit a positive effect on the patient. Nevertheless, the surgeons suggested that it is applicable in the state where the surgical margins are positive or there is a doubt after the excision [18]. There is a reduction of local recurrence in 44% observed in a patient who undergoes radiotherapy [20].

Chemotherapy is another choice for adjuvant therapy required to treat distant metastasis. As in current case following the surgical excision, lymph node dissection and chemotherapy are necessary to treat the metastatic lesions [21]. Before taking the chemotherapy into consideration it is essential to take certain factors into accounts such as age, performance status, size, grade, location, type of initial surgery, and margin status [19,22]. Studies have displayed a positive outcome when both radiotherapy and chemotherapy are used together [17,21]. However, due to the toxic effects these modalities are recommended only to be used in certain cases. Initially after every 4-6 months regular followup is recommended for consecutive 3 years followed by annual check-up [11].

Despite the likelihood of recurrence, the radical orchiectomy with complete margin removal reduces the morbidity and present with a good prognosis. Tumor resection depending upon tumour size and absence of metastasis also provides a satisfactory prognosis if the tumor was removed through wide resection [5]. One of the case reports pointed out that the patient with Poorly-differentiated Myxoidliposarcoma (PDML) suffered a relapse after 6 months. The patient underwent a brief sessions of chemotherapy; however, he couldn't survive. It is reported that well differentiated lipomas and myxoid

lipomas present with a low chance of recurrence and better outcomes.

Researchers have performed tumor staging based upon histological examination, grading and metastasis [23]. According to WHO, soft tissue tumors (liposarcoma) is categorised into five categories: Myxoid (most common), well-differentiated (adipocytic, sclerosing and inflammatory subtypes), dedifferentiated, round-cell and pleomorphic [23]. Low grade subtypes are well differentiated with slight local invasion. High grade lesions occur rarely; nevertheless, has increased incidence of metastatic lesions and recurrence. This high grad tumor often spreads through the hematogenous pathway leading to distant metastases to the bone and lungs. A literature state the infection spreading route does not depends upon the lymphatic system; therefore, removal of lymph nodes does not increase survival chances [16]. Fifty percent of cases show recurrence; however, the average time period for local recurrence includes 40.9 months after surgical treatment [16]. Tan et al. [12], in his study failed to provide a significant relation between recurrence and margin status, tumor size, or tumor grade ($P>0.05$) despite the study struggled at certain points to detect the valid difference. The reports after the procedure exhibited a wide and complete resection with clear microscopic margins indicating successful management of spermatic cord liposarcomas. Therefore, the literature suggests to avoid future recurrence, any sign of positive margin should be removed.

Conclusion

Summarising, liposarcomas are challenging to diagnosis clinically. With no gold standard treatment, the management of the liposarcoma was conducted on the guidance of case reports in the literature. Therefore, it is suggestive that the key to successful treatment depends upon complete resection of the tumor with negative margins. In certain cases where there is a doubt for a positive margin, adjuvant therapies such as radiation and chemo therapy is suggested to elude local recurrence. Lymph node involvement is suggestive of lymph node dissection. Further investigation is necessary for proper guidelines.

References

1. Demirci U, Buyukberber S, Cakir A, Ozturk B, Akyurek N, et al. (2010) Synchronous testicular liposarcoma and prostate adenocarcinoma: a case report. Cases journal. 3(1):27.
2. Montgomery E, Fisher C. (2003) Paratesticular liposarcoma: a clinicopathologic study. AM J SURG PATHOL. 27(1):40-7.
3. Papageorgiou M, Dadakas G, Donev K. (2011)Liposarcoma of the spermatic cord: a case report. J Med Case Rep. 2011.
4. Chalouhy C, Ruck JM, Moukarzel M, Jourdi R, Dagher N,et al. (2017) Current management of liposarcoma of the spermatic cord: A case report and review of the literature. Mol Clinic Oncol. 6(3):438-40.
5. Vinayagam K, Hosamath V, Honnappa S, Rau AR. (2014) Paratesticular liposarcoma-masquerading as a testicular tumour. JCDR. 8(2):165.
6. Li F, Tian R, Yin C, Dai X, Wang H, et al. (2013) Liposarcoma of the spermatic cord mimicking a left inguinal hernia: a case report and literature review. World J Surg Oncol. 11(1):18.
7. Patel NG, Rajagopalan A, Shrotri NS. (2011) Scrotal liposarcoma-a rare extratesticular tumour. JRSM short reports. 2(12):1-3.

8. Parker III RA, Menias CO, Quazi R, Hara AK, Verma S, et al. (2015) MR imaging of the penis and scrotum. Radiographics. 35(4):1033-50.
9. Pollack SM, Loggers ET, Rodler ET, Yee C, Jones RL. (2011) Immune-based therapies for sarcoma. Sarcoma. 2011.
10. De Zordo T, Stronegger D, Pallwein-Prettner L, Harvey CJ, Pinggera G, et al. (2013) Multiparametric ultrasonography of the testicles. Nat Rev Urol. 10(3):135.
11. Pergel A, Yucel AF, Aydin I, Sahin DA, Gucer H, et al. (2010) Paratesticular liposarcoma: a radiologic pathologic correlation. JCIS. 1.
12. Tan C, Dasari B, Smyth J, Brown R. (2011) Liposarcoma of the spermatic cord: a report of two cases. Ann R Coll Surg Engl. 94(1):e10-e2.
13. Parenti GC, Feletti F, Carnevale A, Uccelli L, Giganti M. (2018) Imaging of the scrotum: beyond sonography. Insights into imaging. 9(2):137-48.
14. ap Dafydd D, Messiou C, Thway K, Strauss DC, Nicol DL, Moskovic E. (2017) Paratesticular sarcoma: typical presentation, imaging features, and clinical challenges. Urology. 100:163-8.
15. Chiodini S, Luciani LG, Cai T, Molinari A, Morelli L, et al. (2015) Unusual case of locally advanced and metastatic paratesticular liposarcoma: A case report and review of the literature. Arch Ital Urol Nefrol Androl. 87(1):87-9.
16. Alyousef H, Osman EM, Gomha MA. (2013) Paratesticular liposarcoma: a case report and review of the literature. Urol Case Rep. 2013.
17. Mullinax JE, Zager JS, Gonzalez RJ. (2015) Current diagnosis and management of retroperitoneal sarcoma. Cancer Control. 18(3):177-87.
18. Giridhar V, Kumar PB, Natarajan K, Hegde P. (2011) Testicular leiomyosarcoma with metastasis. Indian journal of urology: IJU. 27(2):278.
19. Di Gregorio M, D'Hondt L, Lorge F, Nollevaux M-C. (2017) Liposarcoma of the spermatic cord: An infrequent pathology. Case Rep Oncol. 10(1):136-42.
20. Hazariwala R, Morris CG, Gilbert S, Algood C, Zlotecki RA. (2013) Radiotherapy for spermatic cord sarcoma. Am J Clin Oncol. 36(4):392-4.
21. Schoonjans C, Servaes D, Bronckaers M. (2015) Liposarcoma scroti: A rare paratesticular tumor. Acta Chirurgica Belgica. 116(2):122-5.
22. Nakamura T, Matsumine A, Matsubara T, Asanuma K, Sudo A. (2016) Neoplastic fever in patients with bone and soft tissue sarcoma. Mol Clin Oncol. 5(5):631-4.
23. Crago AM, Singer S. (2011) Clinical and molecular approaches to well-differentiated and dedifferentiated liposarcoma. CURR OPIN ONCOL. 23(4):373.