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Case report

Paratesticular leiomyoma mimicking an inguinal hernia: A rare case report and literature review

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ABSTRACT

Leiomyomas are considered as a rare, benign, slow-growing, and smooth muscle tumours which may present in all regions in the body. The presentation of leiomyoma in genitourinary tract specifically in paratesticular region is extremely rare. The patients may present with palpable, and painful mass in the inguinal region hence mimicking the nature of inguinal hernia. Herewith, we report our experience of 36-year-old male which had been referred for the suspicion of incarcerated inguinal hernia manifesting with painful mass in the right inguinal and testicular region. During testicular exploration surgery, a benign tumour was incidentally discovered. The lesions were then surgically removed by performing radical orchiectomy with the pathology result confirmed the presence of paratesticular leiomyoma. The post-operative course was uneventful and the patient was discharged on first post-operative day. The patient remained free from metastases or local recurrence after 12 months of regular follow up. This case report demonstrated a rare presentation of paratesticular leiomyoma with misleading manifestation of inguinal hernia. Careful and tailored investigation should be performed to avoid misled diagnosis of this case.

1. Introduction and importance

Anatomically, scrotal lesion consisted of intratesticular and paratesticular pathologies [1]. Paratesticular tumours are benign tumours with several histopathological types, including lipoma, adenomatous, leiomyoma, fibroma, hemangioma, neurofibroma, and cystadenoma [2]. The histological type of leiomyoma tumours are exceedingly rare which only five cases of urinary tract leiomyoma tumours have been reported in Southeast Asia [3]. The overall incidence of paratesticular leiomyomas was up to 17.7 % in real world [4]. Leiomyoma was originated from smooth muscle and may be found in various organ regions [5]. Most patients present with asymptomatic, slow-growing mass and are found incidentally. Moreover, the presentation of painful mass was considered to be rare. Paratesticular tumours may occur at any age and have a 3 % chance of malignancy. The treatment recommendation for the tumour is open surgery along with histopathological confirmation [5]. This work has been reported in line with the SCARE and PROCESS criteria [6,7]. Herewith, we report a case of solitary paratesticular leiomyoma presenting with painful mass with misleading diagnosis of incarcerated inguinal hernia. This report will describe further details on the clinical feature and management of paratesticular leiomyoma.

2. Case presentation

A 36-year-old male patient was referred to our institution with the suspicion of right incarcerated inguinal hernia. The chief complaint was right-sided testicular swelling and pain since 1-day prior admission. The mass was palpable and extremely painful. The mass in the inguinal and testicle emerged for two years and gradually increasing. The patient revealed that the mass sometimes disappeared. There were no history of fever or trauma. On physical examination, a 15 × 10 cm sized tumour was palpable in the right testicle extending into the inguinal region with firm consistency. The left testicle was found normal as shown in Fig. 1. The serum tumour markers examination of human chorionic gonadotropin, lactate dehydrogenase and alpha-fetoprotein were within normal limit. The chest x-ray revealed cardiomegaly, and the KUB image revealed no stones in the urinary system. Abdominal ultrasound examination indicated the presence of mass in the Mc Burney area which was previously identified as intestine as shown in Fig. 2.

Surgical exploration of the testicle through the inguinal region was then performed. The mass was identified to be originated from the paratesticular region which is not inside the testicle and was not identified as hernia. However, due to the tumour was not distinguishable

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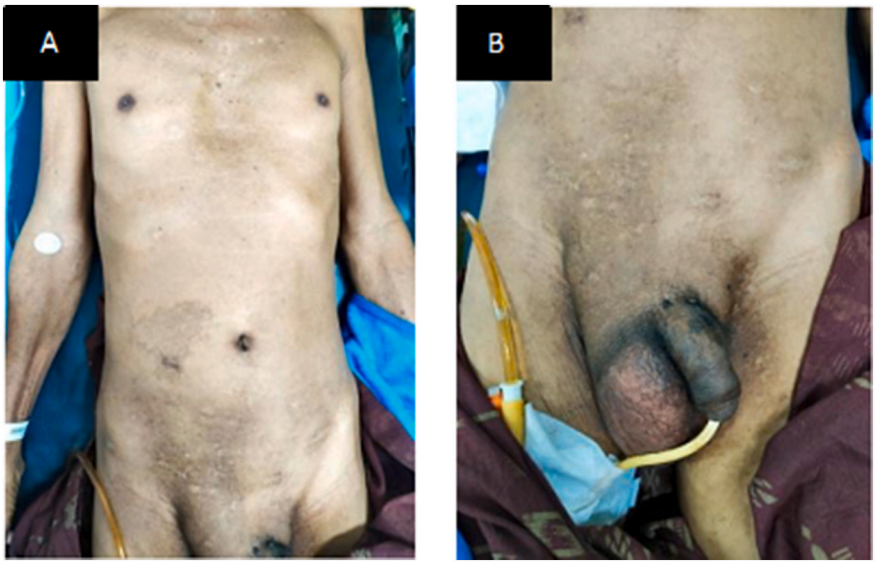

Fig. 1. Clinical appearance showing the presentation of inguinal hernia with right testicular mass.

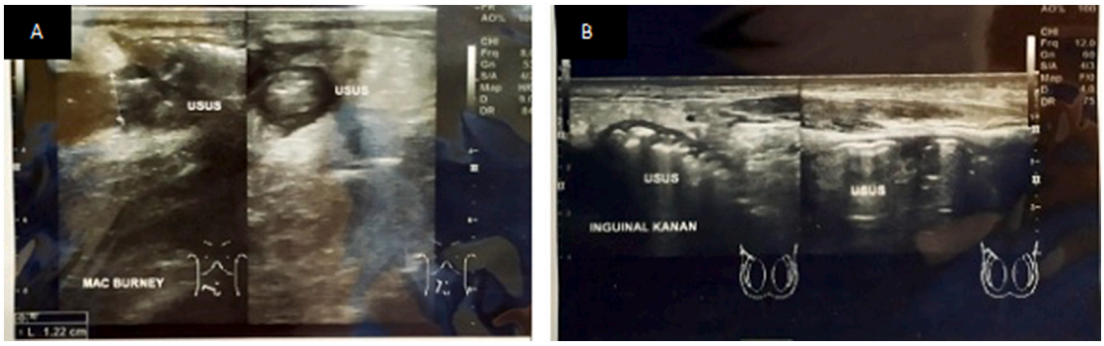

Fig. 2. Ultrasound examination of the abdomen (A) and testicle (B) with suspicion of intestine in the Mcburney area and inside the scrotum.

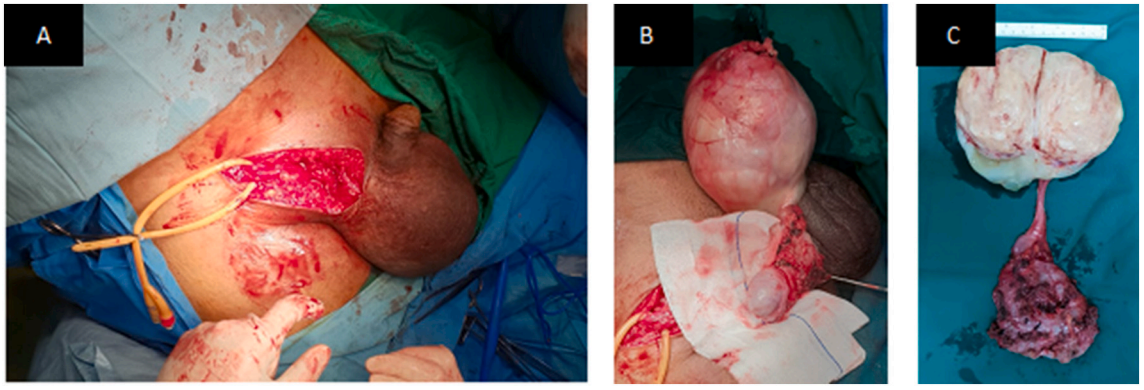

Fig. 3. Right testicular mas was revealed during surgery (A and B), Gross feature of the lesion removed from the scrotum (C).

from the testicle and there was a suspicion of testicular malignancy, radical orchiectomy was performed. Fig. 3 showed the surgical procedure of this case.

The post-operative course was uneventful, patient was stable and was immediately discharged on the first postoperative day. For further examination, the resected mass underwent histopathological examination. The presence of a mesenchymal spindle tumour on pathology confirmed the diagnosis of paratesticular leiomyoma as shown in Fig. 4. The patient was regularly monitored for the last 12 months. The patient remained free from metastases or local recurrence.

3. Clinical discussion

The paratesticular area is a convoluted anatomical region consisted of spermatic cord and its contents including tunica layer, the epididymis, and testicular appendices [8]. The testicle was not in part of paratesticular region. Tumours in paratesticular region may arise from mesenchymal, epithelial, or mesothelial cells [9]. Leiomyomas in the

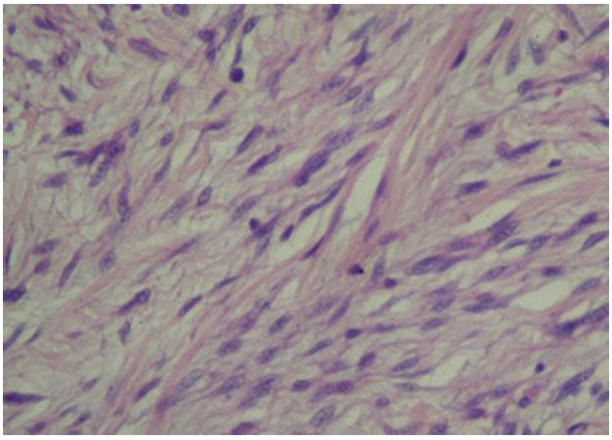

Fig. 4. Histopathological appearance of the right testicular mass.

paratesticular region are very rare, with only a few cases reported [10]. Leiomyomas are mesenchymal cell-derived benign encapsulated tumours which may occur in any region in the genitourinary system as long as the organ contains smooth muscle cell [11]. Scrotal leiomyomas are most commonly occurred in the epididymis, but this case may also be found in the spermatic cord, dartos layer, tunica albuginea, testicular parenchyma and the superficial subcutaneous smooth muscle from the scrotum [12].

Leiomyomas are most frequently reported between the ages of 40 and 50 [13]. In relation to our case, this patient was 36 years old male which is slightly younger than the common prevalence. Previous study has documented a case of a 13-year-old kid with paratesticular leiomyoma, in which this case was considered as one of the youngest case ever documented [14]. The most common symptom of paratesticular leiomyoma is a non-tender mass in the scrotum. This mass may be accompanied by pain in the scrotum above the testicle or in the inguinal area. When the tumour is asymptomatic and grows bigger slowly, most of the patients delayed to seek for treatment until the tumour becomes very big causing bad cosmesis and inducing extreme painful in the mass [15]. In relation to our case, the patient has been complaining the scrotal mass for two years which become very painful lately. Misleading manifestation of paratesticular leiomyoma presenting as incarcerated inguinal hernia is a very rare finding in this case.

Thorough investigation prior surgery should be performed in atypical inguinal masses [16]. Currently, there are no available pathognomonic features to differentiate between specific diagnoses of these tumours [17]. While surgery is the primary therapy, a scrotal USG (Ultrasound), CDU (Colour Doppler Ultrasound), or MRI (Magnetic Resonance Imaging) may be used to confirm the diagnosis. Ultrasound is the first-line imaging modality in patients with suspected scrotal masses to evaluate the mass's size, texture, and boundary, however this imaging modality is operator dependent [15]. Ultrasound may assist in the differentiation between intratesticular or extratesticular masses however to distinguish between benign from malignant tumours is still not feasible [18]. Scrotal CDU may provide information on the location, structure, cystic or solid nature, and degree of vascularization of the lesion. Scrotal MRI may be utilized to rule out malignancy or tumour spread into surrounding tissues, as well as to evaluate more about the tumour. MRI may also be utilized to assist in the differential diagnosis [14]. In relation to this case, ultrasound was performed to evaluate the mass. The result suggested the presence of mass in the Mcburney area which was identified as intestine.

Using a more sensitive and precise imaging modality, on the other hand, is not always essential. Only surgery will facilitate a definitive diagnosis [19]. An inguinal technique should be used to examine paratesticular leiomyoma, which requires substantial spermatic cord and testicular resection and high cord ligation [20]. We then performed a testicular exploration via the inguinal approach due to discomfort and the possibility of strangulated lateral inguinal hernia in this case. Interestingly, instead of inguinal hernia, paratesticular mass was found which later was identified as leiomyomas. In contrast, leiomyoma may be treated conservatively with a simple excision, eliminating the necessity for a major orchiectomy in a young man with future reproductive potential [10]. Moreover, leiomyomas are typically well-defined masses with a white or grey capsule, therefore surgical local excision is a main method for the treatment option [21]. Radical orchiectomy may be performed if there was suspicion of testicular malignancy or when the tumour is not dissociable from the testicle [2,22]. However, in this case, the mass was already contiguous with testicle with suspicion of testicular malignancy. It was also difficult to distinguish the boundary between the tumour and testicle therefore radical orchiectomy was inevitable in this case.

Histopathological examination in leiomyoma may reveal the presence of mature smooth cells arranged in fascicles separated by collagenized stroma with the absence of mitotic activity and coagulation process. The tumour is composed of smooth muscle spindle cells arranged in interlacing bundles with varying amounts of fibrous and hyalinized connective tissue under the microscope [1]. Pathologically, these tumours are classified into four categories based on four characteristics. They include the following: (i) a maximum diameter of more than 5 cm; (ii) an infiltrating margin; (iii) more than five mitotic figures per high-power field; and (IV) significant cytological atypia. In this case, the presence of mesenchymal spindle tumour confirmed the diagnosis of paratesticular leiomyoma.

Following the excision of the mass lesion, an intraoperative frozen section may be performed to rule out malignancies. Positive immunohistochemical staining for SMA, caldesmon, and desmin is required to establish the diagnosis of leiomyoma. Desmin deficiency is associated with an inflammatory myofibroblastic tumour (IMT) of the spermatic cord [21]. Mouse double minute 2 (MDM2) amplification is the hallmark of leiomyosarcoma thus this test was important to rule out the differential diagnosis of leiomyosarcoma. However, immunohistochemistry was not possible in this case due to a lack of resources. It is critical to follow up, and any recurrence should be documented to rule out the possibility of malignancy.

4. Conclusion

Paratesticular leiomyomas are extremely rare benign tumours that may present similarly with an inguinal hernia. Paratesticular leiomyomas should be investigated as a differential diagnosis in men with atypical inguinal mass. Thorough clinical examination, imaging modalities, histopathology and immunohistochemistry may aid in the diagnosis and to avoid the misleading presentation of this disease. Surgical exploration is the most-commonly used treatment. Radical orchiectomy may be performed if there was a sign of testicular malignancy or when the tumour is not distinguishable from the testicle or surrounding tissue.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Ethical approval

Ethical approval has been acquired in this study.

Consent

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Author contribution

Conceptualization – Conceptualization – DI, DMS; Data curation – DI, YUA, IAR; Materials – DI, DMS; Formal Analysis – DI, YUA, IAR; Investigation – DI, YUA, IAR; Methodology – DI, YUA, IAR; Supervision – DMS; Writing original draft – DI, YUA, IAR; Writing, review and editing – DI, IAR, DMS.

Registration of research studies

None.

Guarantor

Doddy M. Soebadi.

Declaration of competing interest

No conflict of interest in this study.

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