

Giant bilateral primary scrotal lipoma along with lipomas in multiple sites of the body: a case report and literature review

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Abstract: Mesenchymal neoplasms of the scrotum are extremely rare in the clinical practices, in which lipoma is the most common benign neoplasm of them. Primary scrotal lipoma refers to the lipoma originated from scrotal wall. We reported a rare case of a 47-year-old man who suffered from bilateral giant primary scrotal lipoma along with lipomas in multiple sites of the body. Scrotal ultrasound indicated that huge hypoechogenic masses were observed in the bilateral scrotums. Contrast-enhanced computed tomography (CT) indicated increased fat density in the scrotal areas. Surgical resection was performed on both scrotal neoplasms. Diagnosis of lipoma was confirmed by the pathological examination through the morphological observation as well as the absence of murine double minute2 (MDM2) gene amplification in the fluorescence in situ hybridization (FISH) test. After five months of follow-up, the wound of the patient recovered well and no sign of local recurrence was observed. Based on the literature review, very few cases of primary scrotal lipoma were reported in the scientific literature up to date, and this is the first report of bilateral primary scrotal lipoma along with multiple lipomas of the body. We presented this case as a rare phenomenon. Although primary scrotal lipoma is rare, clinicians should take it into account when encountering similar scrotal lesions and know the methods for diagnosis and how to make differential diagnosis with other diseases, which is associated with the patient's treatment strategy and prognosis.

Keywords: Primary scrotal lipoma; treatment; diagnosis; case report

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Introduction

Lipoma is the most common benign neoplasm in the scrotal mesenchymal neoplasms which is extremely rare in the clinical practice. Once intrascrotal lesion is detected, lipoma should be taken into account. Scrotal lipoma always originates from the spermatic cord, however, in some rare cases, it can originate outside the spermatic cord such as the scrotal wall (1,2). This article reported a case of a giant bilateral primary scrotal lipoma diagnosed by pathological

examination via surgical resection in a 47-year-old man referred to our department along with lipomas in multiple sites of the body. Based on our literature review, very few cases of primary scrotal lipomas were reported in the scientific literature especially along with multiple lipomas in multiple sites of the body, so we regard this case as a rare one. We present the following article in accordance with the CARE reporting checklist (3) (available at http://dx.doi. org/10.21037/tau-20-1073).

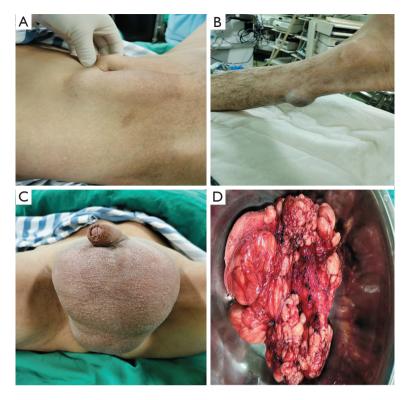


Figure 1 Scrotal lipoma with part of lipomas in multiple sites of the body and surgical specimen. (A) Lipoma in peripheral umbilicus. (B) Lipoma in left Achilles tendon. (C) Giant bilateral scrotal lipoma. (D) Surgical specimen after surgical excision.

Case presentation

A 47 years old man was admitted to our hospital due to the progressively enlarging lumps which was found accidentally in the bilateral scrotums since 8 months ago. These lumps were painless and movable, whose size increased gradually. In particular, the patient found similar lumps occurred in cervical occipital, pectoral back, peripheral umbilicus and left Achilles tendon successively. No significant positive history was provided to help speculate the pathogenesis of the lumps. A comprehensive and systematic physical examination was conducted when the patient was admitted, which showed the results that the lumps in the scrotums were approximately 10 cm × 5 cm in both sides, the cervical occipital was 12 cm × 9 cm, the pectoral back was multiple and variable-sized, the peripheral umbilicus was 10 cm × 8 cm and the left Achilles tendon was $2.5 \text{ cm} \times 2 \text{ cm}$ (Figure 1). The lumps appeared to be painless, non-tender, movable and soft with clear margins. Both testes were not invaded.

A series of examinations were arranged for the patient in order to clarify the diagnosis. The scrotal ultrasound revealed the existence of huge hypoechogenic masses in the bilateral scrotums, the mass size of the right side was about 9.9 cm × 4.5 cm, and the left side was about 10.8 cm × 5.6 cm. Both sides could be deformed by compression with little punctate blood flow signal inside. The testis whose shape and size were not aberrant was pushed into the bilateral inguinal area. Fat-derived tumors were suspected. Contrast-enhanced computed tomography (CT) results indicated the increased fat density in submandibular space, cervical root, item subcutaneous and both scrotal areas. Fat accumulation or lipoma was taken into account (*Figure 2*).

After the comprehensive preoperative preparation, the patient underwent surgery resection under general anesthesia. A 12 cm scrotal vertical incision was given, both sides of the scrotum were enlarged, and there were multiple lobulated large lipomas with complete capsule in the scrotums. The masses were soft, yellow, and irregular in shape. These masses were covered with capsules and slightly adhered to the surrounding tissues (*Figure 1D*). The lipomas were separated from the testes completely. The incision was sewed up after complete hemostasis. No adverse event was observed during the perioperative period. The pathological

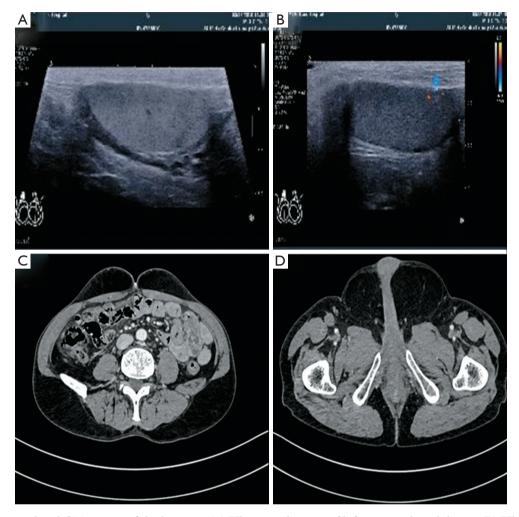


Figure 2 Ultrasound and CT images of the lipomas. (A) Ultrasound image of left testis and epididymis. (B) Ultrasound image of right testis and epididymis. (C) CT image of umbilical lipoma. (D) CT image of scrotal lipoma.

examination indicated a fat-derived tumor. The absence of MDM2 gene was confirmed by FISH test, thus identified the diagnosis of lipoma combined with morphological observation. After five months of follow-up, the wound of the patient recovered well and no recurrence was observed (*Figure 3*). All procedures performed in studies involving human participants were in accordance with the ethical standards of the Ethics committee on biomedical research, West China Hospital of Sichuan University and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

Discussion

The majority of the tumors arised from the extratesticular

mesenchymal soft tissues are benign tumors, including lipoma, leiomyoma and fibrous pseudotumor. Lipoma is the most common benign neoplasm of the scrotum (4). Fujimura *et al.* proposed a classification of scrotal lipomas in consideration of the site of origin: (I) Scrotal lipoma: originated in the spermatic cord with spread to the scrotum. (II) Spermatic cord and tunica vaginalis tumor: originated and developed within the cord. (III) Primary scrotal lipoma: originated within the scrotum (5). According to the classification, scrotal lipoma of this case should be classified as a primary scrotal lipoma.

We conducted a literature search extensively on authenticated databases including PubMed/Medline, Embase, and Web of Science. MeSH terms 'Scrotum' and 'Lipoma' were used as searching strategy, obvious

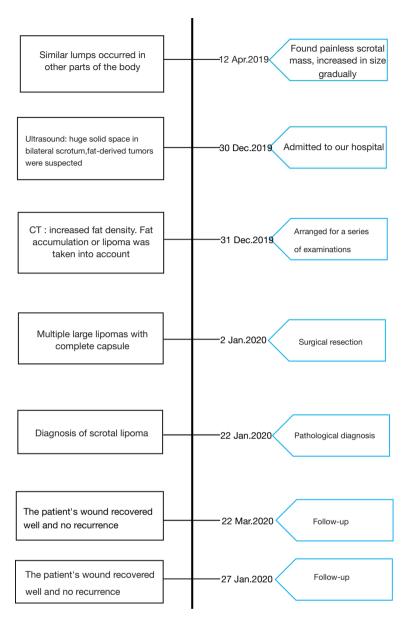


Figure 3 Timeline of the treatment procedure

duplicates were removed, and cases with inaccessible full texts were excluded. Initially, we included all of the cases reported in English with full texts (*Table 1*). The age of scrotal lipoma patients ranged from 1 month to 76 years, the mean age was 45.7 years and the median age was 49 years. It was more commonly occurred on the left scrotum (7/16, 43.75%) rather than the others, while bilateral scrotums involved were quite rare (1/16, 6.25%). According to our literature review, very few cases of primary scrotal lipomas were reported in the scientific literature.

Since the first report by Livermore *et al.* in 1948 (6), only 16 cases of scrotal lipoma have been reported until now, and this is the first report of bilateral primary scrotal lipoma along with multiple lipomas of the body up to date. However, this case needs to be distinguished from Madelung's disease which is a rare benign lesion caused by abnormal metabolism of adipose tissue which has characteristics of excessive, progressive, and symmetrical deposits of adipose tissue in the subcutaneous layer (17). Madelung disease usually occurs in the head and neck, upper trunk, and

Table 1 Patients' information of the case reports of literature review

No. (ref.).	Authors (year)	Age (years)	Place	Clinical manifestation	Ways to diagnose	Therapy	Follow-up (months)	Outcome/ prognosis
1, (6)	Livermore et al. (1948)	75	Left side	Painless mass, sense of weight and pressure, increased in size	Pathology	Surgical resection	N/A	N/A
2, (7)	Cullough et al. (1953)	49	Left side	Painless mass, uncomfortable, sense of pressure	Physical examination, Pathology	Surgical resection	N/A	N/A
3, (8)	Florante et al. (1976)	20	Right side	Painless mass	Physical examination, Pathology	Surgical resection	N/A	Uneventful
4, (8)	Florante et al. (1976)	76	Right side	Painless mass	Physical examination, Pathology	Surgical resection	N/A	N/A
5, (5)	Fujimura et al. (1979)	19	Right side	Painless mass, increased in size	Physical examination, Pathology	Surgical resection	N/A	Uneventful
6, (9)	Rosenberg et al. (1989)	37	Left side	N/A	Ultrasound, Pathology	Surgical resection	N/A	N/A
7, (9)	Rosenberg et al. (1989)	N/A	N/A	Scrotal swelling	Ultrasonography, CT	Surgical resection	N/A	N/A
8, (10)	Szmigielski et al. (2000)	67	Both sides	Scrotal swelling	Physical examination, Ultrasonography, CT, Pathology	Surgical resection	N/A	N/A
9 (11)	Edelstein et al. (2009)	48	N/A	Scrotal swelling	Pathology	Surgical resection	N/A	N/A
10, (12)	Kim <i>et al.</i> (2009)	1 month	Midline	Pendulous scrotal mass, increased in size	Ultrasound, Pathology	Surgical resection	N/A	N/A
11, (13)	Sakorafas et al. (2010)	67	N/A	Mild discomfort	Physical examination, Ultrasonography, MRI, Pathology	Surgical resection	N/A	Uneventful
12, (14)	Kaplanoglu et al. (2013)	64	Left side	Painless mass, swelling	Physical examination, Ultrasonography, Pathology	Surgical resection	Six	No recurrence
13, (1)	Fabiani et al. (2016)	22	Left side	Painful mass	Physical examination, Ultrasonography, Pathology	Surgical resection	N/A	N/A
14, (15)	Creta et al. (2017)	54	Midline	Scrotal swelling, discomfort	Physical examination, Ultrasonography, MRI, Pathology	Surgical resection	N/A	N/A
15, (2)	Srivastava et al. (2017)	29	Left side	Painless mass, increased in size.	Physical examination, Ultrasonography, MRI, Pathology	Surgical resection	N/A	Uneventful
16, (16)	Yamamichi et al. (2018)	58	Left side	Scrotal swelling, discomfort	CT, MRI, PET-CT, Pathology	Surgical resection	Six	No recurrence

N/A, not available; CT, computed tomography; MRI, magnetic resonance imaging; PET-CT, positron emission tomography-computed tomography.

proximal portions of the upper extremities, specific and rare sites such as scrotal involvement associated with bilateral asymmetrical gynaecomastia were also reported previously, which seemed quite similar to the case in this study. Lipoma also has similar imaging manifestations to the Madelung disease, while the key to distinguish lipoma from fatty non-encapsulated tissue of Madelung disease is the complete capsules of the lipoma (18). In our case, the surgical specimen of the patient had complete capsules, proving that it was lipoma.

In addition to the comprehensive and systematic physical examination, ultrasonography and imaging examinations play critical roles in the evaluation of scrotal masses. Ultrasonography is recommended as one of the firstline examinations in the identification of lipomas due to its high feasibility as well as the ability to distinguish cystic or solid lesions under both intra- or extra-testicular conditions. Lipoma is usually characterized as a welldefined, ovalar, sometimes capsulated and "striped" mass in ultrasound images, which lacks internal blood flow. However, ultrasound findings are often nonspecific and may be variable (4,19). CT can detect subtle density differences and make anatomic assessment accurately. The unique tissue density of lipomatous masses is an obvious character that can be used to distinguish them from other soft tissue tumors. Lipomas, avascular at angiography, demonstrate a homogeneous, sharply marginated low density mass on CT scan, whereas liposarcoma with heterogeneous mixture of fat and water density presents heterogeneity, ill-defined margins, and contrast enhancement. However, it is still difficult to make a differential diagnosis with liposarcoma by CT in some cases. For example, the angiolipoma has similar features to the liposarcoma, which made it difficult to distinguish angiolipoma from liposarcoma although angiolipoma appears no definite invasion on the CT scan (20). Lipoma is similar to subcutaneous fat but may contain little thin septa and demonstrate a characterized appearance at magnetic resonance imaging (MRI) (21). It can be differentiated from liposarcoma by observing the absence of enhancement after administration of gadolinium contrast material. Imaging findings can often help urologists for both patient counseling and surgical planning (2,4). Although the MRI imaging is sensitive, in some cases, however, there may be few non adipose components in a large lipoma such as blood vessels, fibrous tissues, and muscle fibers similar to a well differentiated liposarcoma, which made the differential diagnosis complicated (14,22).

Symptoms vary with size and pressure caused by the

lipoma. Our literature review results revealed that the most common symptoms were painless mass of the scrotum (7/16, 43.75%) and scrotal swelling or discomfort (7/16, 43.75%), followed by sense of weight and pressure (2/16, 12.50%) and painful mass (1/16, 6.25%). The patient in our case presented with painless mass of bilateral scrotums. All of the patients underwent surgical resection, the postoperative course was uneventful and no recurrence was observed after six months follow-up in two cases. Surgical resection remains predominant therapeutic regimen in the first-line treatment of a primary scrotal lipoma, simple enucleation through scrotal or combined scrotal and inguinal incision will suffice (2). Histopathology is always essential for diagnosis, sometimes genetic testing is needed for differential diagnosis in some cases. Amplification of MDM2 and cyclin-dependent kinase 4 (CDK4) genes are the most prominent genetic aberrations of ALTs and dedifferentiated liposarcoma (DDLPS). MDM2 gene amplification is a more reproducible and reliable method to diagnose DDLPS, the sensitivity and specificity of which was 100% and 83.3% respectively (23). In this case, the pathological examination indicated a fat-derived tumor and FISH test did not show amplification of MDM2 gene, the diagnosis of lipoma was confirmed combined with morphology.

In this study, a rare case of bilateral primary scrotal lipoma along with lipomas in multiple sites of the body is reported. Literature review was also conducted to summarize the features of scrotal lipoma, which could provide clinicians with ideas and methods for diagnosis when encountering such scrotal lesions. Meanwhile, we had proposed the differential diagnosis between scrotal lipoma and Madelung's disease, which was very similar to our case and needed to be paid attention to in clinical practices. Our limitations were that: (I) MRI was not performed in this patient, since MRI needed a relatively long time to make an appointment, while the patient required surgery resection as soon as possible. (II) Lipomas in other parts of the body confirmed by CT were symptomless and had no impact on the patient, the patient had no intention to remove them, so the pathological examination of lipomas in other parts were not available.

Conclusions

Primary scrotal lipomas are rare, benign paratesticular tumors, and pose significant diagnostic dilemma. Ultrasonography is one of the first-line examinations, and imaging examinations also play critical roles in the evaluation of scrotal masses. Histopathology and genetic testing are always essential for a precise diagnosis. Surgical resection remains predominant treatment in the first-line management of scrotal lipomas which can solve oppression symptoms and provide histopathologic diagnosis.

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Footnote

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the Ethics committee on biomedical research, West China Hospital of Sichuan University and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient.

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