

World Journal of *Clinical Cases*

World J Clin Cases 2022 October 16; 10(29): 10391-10822



STANDARD AND CONSENSUS

- 10391** Baishideng's *Reference Citation Analysis* database announces the first *Article Influence Index* of multidisciplinary scholars
Wang JL, Ma YJ, Ma L, Ma N, Guo DM, Ma LS

REVIEW

- 10399** Cholecystectomy for asymptomatic gallstones: Markov decision tree analysis
Lee BJH, Yap QV, Low JK, Chan YH, Shelat VG
- 10413** Liver transplantation for hepatocellular carcinoma: Historical evolution of transplantation criteria
Ince V, Sahin TT, Akbulut S, Yilmaz S

MINIREVIEWS

- 10428** Prostate only radiotherapy using external beam radiotherapy: A clinician's perspective
Lee JW, Chung MJ

ORIGINAL ARTICLE**Retrospective Study**

- 10435** Age-adjusted NT-proBNP could help in the early identification and follow-up of children at risk for severe multisystem inflammatory syndrome associated with COVID-19 (MIS-C)
Rodriguez-Gonzalez M, Castellano-Martinez A
- 10451** Clinicopathological characteristics and prognosis of gastric signet ring cell carcinoma
Tian HK, Zhang Z, Ning ZK, Liu J, Liu ZT, Huang HY, Zong Z, Li H
- 10467** Development and validation of a prognostic nomogram for decompensated liver cirrhosis
Zhang W, Zhang Y, Liu Q, Nie Y, Zhu X

Observational Study

- 10478** Effect of medical care linkage-continuous management mode in patients with posterior circulation cerebral infarction undergoing endovascular interventional therapy
Zhu FX, Ye Q
- 10487** Effect of the COVID-19 pandemic on patients with presumed diagnosis of acute appendicitis
Akbulut S, Tuncer A, Ogut Z, Sahin TT, Koc C, Guldogan E, Karabulut E, Tanriverdi ES, Ozer A

EVIDENCE-BASED MEDICINE

- 10501** Delineation of a SMARCA4-specific competing endogenous RNA network and its function in hepatocellular carcinoma

Zhang L, Sun T, Wu XY, Fei FM, Gao ZZ

SYSTEMATIC REVIEWS

- 10516** Comparison of laboratory parameters, clinical symptoms and clinical outcomes of COVID-19 and influenza in pediatric patients: A systematic review and meta-analysis

Yu B, Chen HH, Hu XF, Mai RZ, He HY

CASE REPORT

- 10529** Surgical treatment of bipolar segmental clavicle fracture: A case report

Liang L, Chen XL, Chen Y, Zhang NN

- 10535** Multiple disciplinary team management of rare primary splenic malignancy: Two case reports

Luo H, Wang T, Xiao L, Wang C, Yi H

- 10543** Klippel-Trenaunay-Weber syndrome with ischemic stroke: A case report

Lee G, Choi T

- 10550** Vedolizumab in the treatment of immune checkpoint inhibitor-induced colitis: Two case reports

Zhang Z, Zheng CQ

- 10559** Novel way of patent foramen ovale detection and percutaneous closure by intracardiac echocardiography: A case report

Han KN, Yang SW, Zhou YJ

- 10565** Treatment failure in a patient infected with *Listeria* sepsis combined with latent meningitis: A case report

Wu GX, Zhou JY, Hong WJ, Huang J, Yan SQ

- 10575** Three-in-one incidence of hepatocellular carcinoma, cholangiocellular carcinoma, and neuroendocrine carcinoma: A case report

Wu Y, Xie CB, He YH, Ke D, Huang Q, Zhao KF, Shi RS

- 10583** Intestinal microbiome changes in an infant with right atrial isomerism and recurrent necrotizing enterocolitis: A case report and review of literature

Kaplina A, Zaikova E, Ivanov A, Volkova Y, Alkhova T, Nikiforov V, Latypov A, Khavkina M, Fedoseeva T, Pervunina T, Skorobogatova Y, Volkova S, Ulyantsev V, Kalinina O, Sitkin S, Petrova N

- 10600** *Serratia fonticola* and its role as a single pathogen causing emphysematous pyelonephritis in a non-diabetic patient: A case report

Villasuso-Alcocer V, Flores-Tapia JP, Perez-Garfias F, Rochel-Perez A, Mendez-Dominguez N

- 10606** Cardiac myxoma shedding leads to lower extremity arterial embolism: A case report

Meng XH, Xie LS, Xie XP, Liu YC, Huang CP, Wang LJ, Zhang GH, Xu D, Cai XC, Fang X

- 10614** Extracorporeal membrane oxygenation in curing a young man after modified Fontan operation: A case report
Guo HB, Tan JB, Cui YC, Xiong HF, Li CS, Liu YF, Sun Y, Pu L, Xiang P, Zhang M, Hao JJ, Yin NN, Hou XT, Liu JY
- 10622** Wandering small intestinal stromal tumor: A case report
Su JZ, Fan SF, Song X, Cao LJ, Su DY
- 10629** Acute mesenteric ischemia secondary to oral contraceptive-induced portomesenteric and splenic vein thrombosis: A case report
Zhao JW, Cui XH, Zhao WY, Wang L, Xing L, Jiang XY, Gong X, Yu L
- 10638** Perioperative anesthesia management in pediatric liver transplant recipient with atrial septal defect: A case report
Liu L, Chen P, Fang LL, Yu LN
- 10647** Multiple tophi deposits in the spine: A case report
Chen HJ, Chen DY, Zhou SZ, Chi KD, Wu JZ, Huang FL
- 10655** Myeloproliferative neoplasms complicated with β -thalassemia: Two case report
Xu NW, Li LJ
- 10663** Synchronous renal pelvis carcinoma associated with small lymphocytic lymphoma: A case report
Yang HJ, Huang X
- 10670** *Leclercia adecarboxylata* infective endocarditis in a man with mitral stenosis: A case report and review of the literature
Tan R, Yu JQ, Wang J, Zheng RQ
- 10681** Progressive ataxia of cerebrotendinous xanthomatosis with a rare c.255+1G>T splice site mutation: A case report
Chang YY, Yu CQ, Zhu L
- 10689** Intravesical explosion during transurethral resection of bladder tumor: A case report
Xu CB, Jia DS, Pan ZS
- 10695** Submucosal esophageal abscess evolving into intramural submucosal dissection: A case report
Jiao Y, Sikong YH, Zhang AJ, Zuo XL, Gao PY, Ren QG, Li RY
- 10701** Immune checkpoint inhibitor-associated arthritis in advanced pulmonary adenocarcinoma: A case report
Yang Y, Huang XJ
- 10708** Chondroid syringoma of the lower back simulating lipoma: A case report
Huang QF, Shao Y, Yu B, Hu XP
- 10713** Tension-reduced closure of large abdominal wall defect caused by shotgun wound: A case report
Li Y, Xing JH, Yang Z, Xu YJ, Yin XY, Chi Y, Xu YC, Han YD, Chen YB, Han Y

- 10721** Myocardial bridging phenomenon is not invariable: A case report
Li HH, Liu MW, Zhang YF, Song BC, Zhu ZC, Zhao FH
- 10728** Recurrent atypical leiomyoma in bladder trigone, confused with uterine fibroids: A case report
Song J, Song H, Kim YW
- 10735** Eczema herpeticum *vs* dermatitis herpetiformis as a clue of dedicator of cytokinesis 8 deficiency diagnosis: A case report
Alshengeti A
- 10742** Cutaneous allergic reaction to subcutaneous vitamin K₁: A case report and review of literature
Zhang M, Chen J, Wang CX, Lin NX, Li X
- 10755** Perithyroidal hemorrhage caused by hydrodissection during radiofrequency ablation for benign thyroid nodules: Two case reports
Zheng BW, Wu T, Yao ZC, Ma YP, Ren J
- 10763** Malignant giant cell tumors of the tendon sheath of the right hip: A case report
Huang WP, Gao G, Yang Q, Chen Z, Qiu YK, Gao JB, Kang L
- 10772** Atypical Takotsubo cardiomyopathy presenting as acute coronary syndrome: A case report
Wang ZH, Fan JR, Zhang GY, Li XL, Li L
- 10779** Secondary light chain amyloidosis with Waldenström's macroglobulinemia and intermodal marginal zone lymphoma: A case report
Zhao ZY, Tang N, Fu XJ, Lin LE
- 10787** Bilateral occurrence of sperm granulomas in the left spermatic cord and on the right epididymis: A case report
Ly DY, Xie HJ, Cui F, Zhou HY, Shuang WB
- 10794** Glucocorticoids combined with tofacitinib in the treatment of Castleman's disease: A case report
Liu XR, Tian M
- 10803** Giant bilateral scrotal lipoma with abnormal somatic fat distribution: A case report
Chen Y, Li XN, Yi XL, Tang Y
- 10811** Elevated procalcitonin levels in the absence of infection in procalcitonin-secreting hepatocellular carcinoma: A case report
Zeng JT, Wang Y, Wang Y, Luo ZH, Qing Z, Zhang Y, Zhang YL, Zhang JF, Li DW, Luo XZ

LETTER TO THE EDITOR

- 10817** "Helicobacter pylori treatment guideline: An Indian perspective": Letter to the editor
Swarnakar R, Yadav SL
- 10820** Effect of gender on the reliability of COVID-19 rapid antigen test among elderly
Nori W, Akram W

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RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Hua-Ge Yu*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lei Wang*.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

October 16, 2022

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INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

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<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Bilateral occurrence of sperm granulomas in the left spermatic cord and on the right epididymis: A case report

Ding-Yang Lv, Hong-Jie Xie, Fan Cui, Hui-Yu Zhou, Wei-Bing Shuang

Specialty type: Andrology

Provenance and peer review:

Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0

Grade B (Very good): 0

Grade C (Good): C, C

Grade D (Fair): 0

Grade E (Poor): 0

P-Reviewer: Arslan M, Turkey;

Exbrayat JM, France

Received: June 28, 2022

Peer-review started: June 28, 2022

First decision: July 14, 2022

Revised: July 22, 2022

Accepted: September 8, 2022

Article in press: September 8, 2022

Published online: October 16, 2022



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Abstract

BACKGROUND

Sperm granuloma is a rare disease in clinical andrology and its incidence is still unclear worldwide. According to the existing literature, sperm granuloma often occurs unilaterally. Clinical and ultrasound features are similar to epididymal tuberculosis, chronic epididymitis and other diseases. Sperm granuloma is usually diagnosed based on postoperative histopathological and immunohistochemical examination.

CASE SUMMARY

A 46-year-old man was admitted to the hospital due to the presence of a left scrotal mass for 3 mo and aggravation of pain for 1 wk. The lesions at both sites were surgically resected. Postoperative pathological examination showed that the left spermatic cord mass and the right epididymal mass were consistent with sperm granuloma. The sperm granulomas then recurred 3 mo after surgery. There is little change in the local mass so far.

CONCLUSION

The case report is helpful for our understanding of this disease. In clinical diagnosis, it should be distinguished from epididymal tuberculosis, chronic epididymitis and other diseases. Color Doppler ultrasound can be used as a preferred examination method but postoperative pathological examination is still needed for diagnosis.

Key Words: Sperm granuloma; Epididymis; Spermatic cord; Recurrence; Case report

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Core Tip: We reported on a rare case of sperm granuloma which occurred bilaterally in the left spermatic cord and on the right epididymis. Remarkably, the granuloma recurred at 3 mo after surgery and this recurrence has not been reported before. Since the patient has no history of vasectomy, tumor or trauma, it is more likely that this case of sperm granuloma is merely caused by inflammation which is rare for sperm granuloma. This case report can greatly enhance our understanding of sperm granuloma and it is helpful for differential diagnosis of various epididymal lesions in clinical practice.

Citation: Lv DY, Xie HJ, Cui F, Zhou HY, Shuang WB. Bilateral occurrence of sperm granulomas in the left spermatic cord and on the right epididymis: A case report. *World J Clin Cases* 2022; 10(29): 10787-10793

URL: <https://www.wjgnet.com/2307-8960/full/v10/i29/10787.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v10.i29.10787>

INTRODUCTION

Sperm granuloma (SG) is a rare disease and its incidence is still unclear worldwide. The first case was reported in 1949[1] and ultrasonographic findings were first described in 1982[2]. In the past 10 years, only a few cases of SGs have been reported[3-5], let alone the cases of bilateral and postoperative recurrent SGs. According to the existing literature, SG is common in the epididymis and occasionally occurs in the spermatic cord and testes. SG is usually seen in unilateral nodules. Most of the patients visited a doctor with complaint of scrotal pain and induration.

Herein, we report a rare type of SG which occurred bilaterally, in both the left spermatic cord and right epididymis. The postoperative pathological diagnosis was consistent with the manifestation of SG and it recurred 3 mo after surgery.

CASE PRESENTATION

Chief complaints

A 46-year-old man reported having found a bean-like mass in the left scrotum approximately 3 mo prior.

History of present illness

The patient described his having felt a bean-like mass in the left scrotum without obvious inducement 3 mo prior. He described the scrotal mass as firm and associated with mild pain but without the scrotal skin having any redness or itching. The patient denied having any other symptoms, such as lower urinary tract symptoms, hot flashes and night sweats, fever and shiver, or progressive emaciation. Over time, the mass had gradually increased in size, accompanied by irregular swelling and pain. Anti-infective treatment did not relieve the symptoms, and ultimately he presented himself to the urology clinic of our hospital.

History of past illness

The patient denied a history of tuberculosis or vasectomy.

Personal and family history

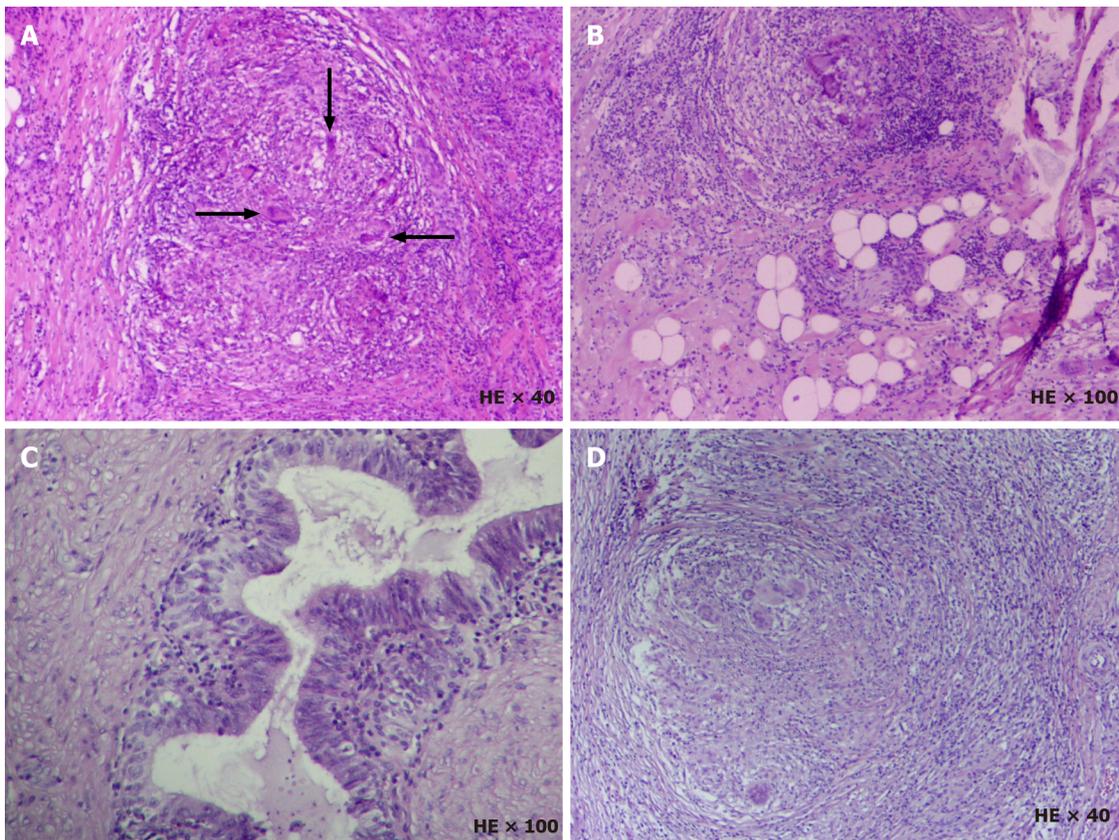
The patient denied any family history of disease of the scrotum.

Physical examination

The physical examination showed a firm mass measuring 4 cm × 3 cm × 2 cm in the left scrotum with no obvious tenderness. The light transmission test was negative.

Laboratory examinations

The mass was negative for three tumor markers including alpha-fetoprotein, human chorionic gonadotropin and lactate dehydrogenase. The surgical specimen was stained with hematoxylin and eosin, and histopathological observation showed that the left spermatic cord mass and the right epididymal mass were consistent with SG (Figure 1). The surgical specimen was processed using indirect immunohistochemistry for CK (Beijing Zhongshanjinqiao Biotechnology Co., Ltd, Beijing, China) and CD68 (Beijing Zhongshanjinqiao Biotechnology Co., Ltd), as well as subjected to acid-fast stain (BaSO Diagnostics Inc., Zhuhai, China); these experiments were performed according to the manufacturer's protocols. The results for the left spermatic cord mass were as follows: acid-fast (-), CK (-) and CD68 (+).



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Figure 1 Histopathological analysis of the resected specimen. A and B: The left scrotum. Spermatic cord mass is consistent with granulomatous lesion with little necrosis. Macrophages can be seen to engulf degraded sperm (arrow), considered as sperm granuloma. The incisional margin did not change significantly. There was no significant change in testis and epididymis; C and D: The right epididymis was infiltrated with lymphocytes, mononuclear cells and eosinophils. The formation of an epithelioid granuloma was found in the epididymis along with multinucleated giant cells, including foreign body giant cells and Langhans giant cells. Coagulative necrosis was found in some areas along with complete necrosis of small focus which is consistent with granulomatous lesions. HE: Hematoxylin-eosin staining.

Imaging examinations

Color Doppler ultrasound (CDU) of the scrotum and positron emission tomography/computed tomography images are shown in [Figure 2](#).

FINAL DIAGNOSIS

Postoperative and intraoperative pathological examination showed that the left spermatic cord mass and the right epididymal mass were consistent with SG.

TREATMENT

To relieve the symptoms, the patient underwent surgery with general anesthesia. During the operation, a fusiform hard mass was seen in the spermatic region of the left proximal epididymis which was about 5.0 cm × 3.0 cm × 2.8 cm in size ([Figure 3A](#) and [B](#)). Because the blood supply to the left testis could not be preserved after the resection of the left spermatic cord tumor, left orchiectomy was performed. The interoperative frozen section analysis indicated inflammatory hyperplasia in the tumor tissue. A hard mass of about 2.0 cm × 1.5 cm × 1.3 cm was found in the tail of the right epididymis and the boundary was still clear. Therefore, the resection of the right epididymis was performed ([Figure 3C](#)). After surgery, the patient was given symptomatic supportive treatment and was discharged after 1 wk.

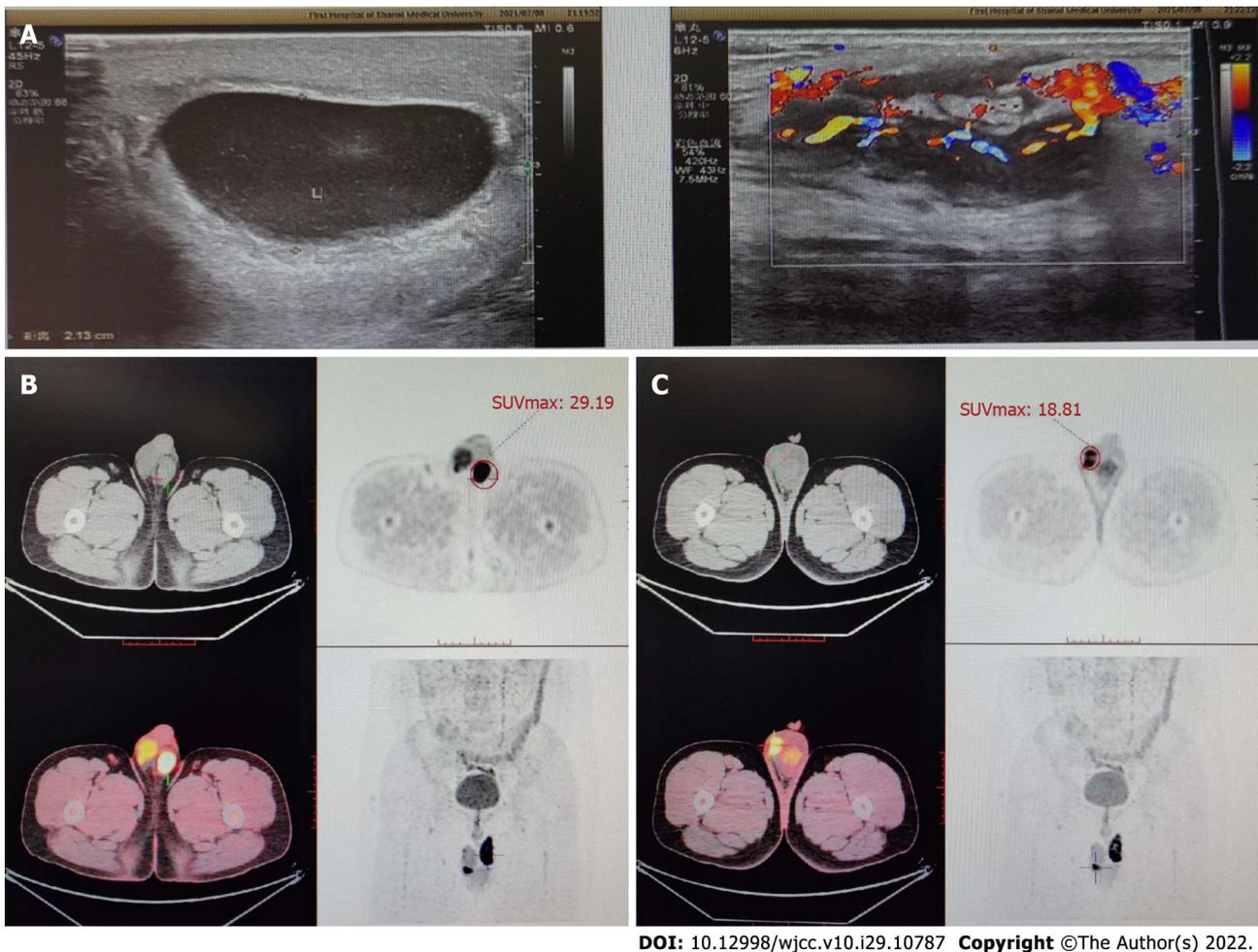


Figure 2 Imaging examinations. A: Color Doppler ultrasound images of the scrotum. There were semi-swirl-like changes in the structure of the left spermatic cord and the echo decreased and was uneven; there was no obvious blood flow signal in the spermatic vein and the blood flow in the artery was filled well; B: Positron emission tomography, the metabolism of the soft tissue nodules of the spermatic vein in the left scrotum increased with a delayed maximum standardized uptake value (SUVmax) of 29.19; C: Computed tomography images of the scrotum, the focal metabolism in the right epididymis increased with a delayed SUVmax of 18.81.

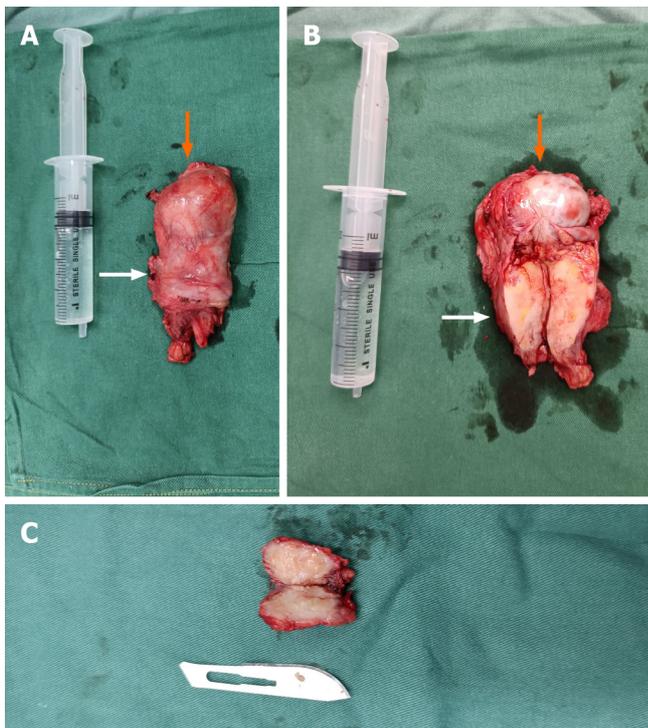
OUTCOME AND FOLLOW-UP

Three months after surgery, the patient went to the outpatient clinic of our hospital once again because of a right scrotal mass. CDU of the scrotum and spermatic cord showed that after left orchietomy, there was no obvious abnormality; the right epididymis was enlarged and the echo of parenchyma was uneven. The color Doppler flow imaging showed an increase in blood flow signals. Considering the possibility of a recurrence of the lesion in the right epididymis, the patient was asked to stay for continuous observation. There is little change in the local mass so far.

DISCUSSION

SG is a chronic granulomatous inflammation with low incidence which is caused by a foreign body reaction with sperm or acid-resistant lipids. Sperm overflow from damaged seminiferous tubules, epididymal ducts or vas deferens to the surrounding stroma can result in the formation of inflammatory granuloma. The breakage of these tubules is often caused by inflammation, tumor, trauma, vasectomy and surgical operation of adjacent sites. Among these various causes, vasectomy is the most common with a rate of epididymal SG after vasectomy of 41% [2]. Nevertheless, there are few reports on SG caused by inflammation, trauma or tumor alone. In this case, the patient had no history of vasectomy, tumor or trauma; therefore, inflammation was the more likely cause. Furthermore, scrotal ultrasound showed that the arterial blood flow was well filled which served as indirect evidence of inflammation.

There are three types of SGs based on their location including testicular SG, epididymal SG and spermatic cord SG [6]. The most common site is epididymis followed by spermatic cord and testis. SG is usually seen in unilateral nodules. In this case, the patient presented with bilateral SGs which occurred in the left spermatic cord and on the right epididymis. This type of case is rare and, likewise, has rarely



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Figure 3 Photograph of the scrotal mass. A: The left testis (orange arrow); B: The left spermatic cord mass (white arrow); C: The mass in the tail of the right epididymis. The size of the syringe is 20 mL.

been reported in the literature.

The diagnosis of SG mainly depends on the pathological examination of surgical specimens. CDU, CT and magnetic resonance imaging all lack specificity. However, CDU can be used as a preferable diagnostic method because CDU images can reveal histopathological characteristics of tumors at different stages[7]. At the early stage (the acute stage), there are a large number of inflammatory cells and sperm cells in the lesion site accompanied by a few fibrous cells; so, the lesions detected by CDU are hypoechoic. In addition, due to inflammatory stimulation, localized hyperemia, edema and vasodilation, there is abundant blood flow in and around the lesion. At the middle stage, inflammatory cells and sperm cells in the stroma gradually decrease but fibrous tissue gradually increases; so, the lesions detected by CDU are moderately hypoechoic. If the fibrous tissue proliferates heterogeneously or tissue begins to necrotize, the echotexture is heterogeneous. As the disease progresses, inflammatory cells and sperm cells further decrease, accompanied by intensive proliferation of fibrous tissue; so, ultrasound shows high echo. During this period, there are few inflammatory cells, and vasodilation and congestion decreases significantly; so, there is only a little stellate blood flow or no blood flow around and inside the focus[7]. In this case, based on the ultrasonographic findings and postoperative pathological examination, the lesion was considered to be in the middle pathological stage.

SGs lack typical clinical features and are often characterized by pain in the scrotum or groin, palpable hard nodules in the epididymis and seminal cord, and sometimes a slight squeezing pain. Therefore, it needs to be differentiated from epididymal tuberculosis, chronic epididymitis, epididymal tumor, epididymal cyst, semen cyst and testicular lesions. Most of the patients with epididymal tuberculosis have a history of tuberculosis, such as afternoon hot flashes, increased erythrocyte sedimentation rate, positive tuberculin test and so on. Ultrasonic images show diffuse enlargement of the epididymis, irregular shape, moderate echo or mixed echo due to calcification[8]. As for the patients with chronic epididymitis, the ultrasonographic examination shows moderate or low echo, and most have a history of acute epididymitis and are improved after anti-infective treatment[9]. With the progression of chronic epididymitis, chronic inflammatory exudates are discharged from local tissues accompanied by tissue edema with cystic changes but no hard nodules detected upon touch. In comparison, SG could be characterized by fibrous tissue hyperplasia, epididymal duct fibrosis and hard nodules near the scrotum [10]. As for epididymal tumors, they are local solid masses and are rarely reported. They can be either benign or malignant. The benign tumor has a clear boundary, with an even low internal echo or moderate-to-high internal echo. The malignant tumor grows rapidly with an uneven internal echo. As for epididymal cyst, though there is no echo in ultrasound examination, CT examination shows low-density shadow due to the presence of fluid which is easily neglected during diagnosis. As for the semen cyst, it was mostly located in the head of the epididymis and appears as round anechoic nodules while SG is mostly located in the tail of the epididymis[11].

SGs are a benign tumor with a very low risk of malignant transformation. As the disease progresses, the lesion may block the vas deferens leading to infertility. Therefore, surgical resection is recommended and should be performed as soon as possible. If the tumor is not completely removed, however, it may recur after surgery.

CONCLUSION

We reported a rare case of SG which occurred bilaterally in the left spermatic cord and on the right epididymis. Remarkably, it recurred at 3 mo after surgery and this recurrence has not been reported before. Since the patient has no history of vasectomy, tumor or trauma, it is more likely that this case of SG is merely caused by inflammation which is rare for SG. This case report can greatly enhance our understanding of SG and it will be helpful in differential diagnosis of various epididymal lesions in clinical practice.

FOOTNOTES

Author contributions: Lv DY contributed to the manuscript writing, editing and data collection; Xie HJ contributed to the data collation; Cui F conducted the literature review; Zhou HY and Shuang WB revised the manuscript; All authors have read and approved the final manuscript.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: All authors declare that they have no conflict of interest to disclose.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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Country/Territory of origin: China

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S-Editor: Liu JH

L-Editor: Filipodia

P-Editor: Liu JH

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