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Case Report

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# Case Report: Postoperative Epididymis Cyst of Transverse Testicular Ectopia with Incarcerated Hernia

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## Abstract

**Background:** Transverse Testicular Ectopia (TTE) is a rare form of urogenital system abnormality, which can be accompanied by inguinal hernia. Currently, there are less than 150 cases reported.

**Case presentation**: The 7 years old male, was admitted to hospital due to "bilateral scrotal masses for six months". Six months ago, the parents found bilateral scrotal enlargement without obvious inducement, further ultrasound proved bilateral cystic masses in the scrotum, and lymphatic malformations were not excluded. Previously, "incarcerated hernia release + orchiopexy" was performed in the neonatal period due to "incarcerated hernia combined with transverse testicular ectopia". During the previous surgery, both testicles were found to be located in the left scrotum, and both testicles showed ischemic changes. One testicle was assessed as necrotic, and the remove of it was highly recommended. But due to the parents' final decision, the necrotic testicle was still being reserved. To our surprise. Regular ultrasound examination after discharge showed that both testicular volumes increased with good blood supply.

**Conclusion**: This case report is helpful to determine whether to choose orchiectomy or conservative orchiopexy to avoid testicular resection for testicular ischemic necrosis caused by the scrotal emergency of neonates. The necrotic testis may still be recovered due to the strong testis ischemia tolerance in neonates. It is very rare to see acquired lymphangioma involving the scrotum. Easily confused with scrotal lymphangioma. Surgical resection is the main treatment.

**Keywords**: Epididymal cyst; Transverse testicular ectopia; Incarcerated hernia; Scrotal lymphangioma

**Abbreviations:** TTE: Transverse Testicular Ectopia

## Background

Transverse Testicular Ectopia (TTE) is a rare form of urogenital system abnormality, which can be accompanied by inguinal hernia. Currently, there are less than 150 cases reported [1-3]. Incarcerated hernia can cause intestinal obstruction, ovarian or testicular ischemia, with an incidence of 6%~31% among children [2]. Both diseases above are scrotal emergency which need emergency operations. Epididymal cyst is a benign fluid-filled sac in the testicles [4]. Easily confused with scrotal lymphangioma.

Surgical resection is the main treatment. We report this case in order to understand the pathogenesis of epididymal cyst and discuss surgical options of testicular ischemia due to incarcerated hernia during the neonatal period.

## Case Presentation

The 7 years old male, was admitted to hospital due to "bilateral scrotal masses for six months" (Figure1).

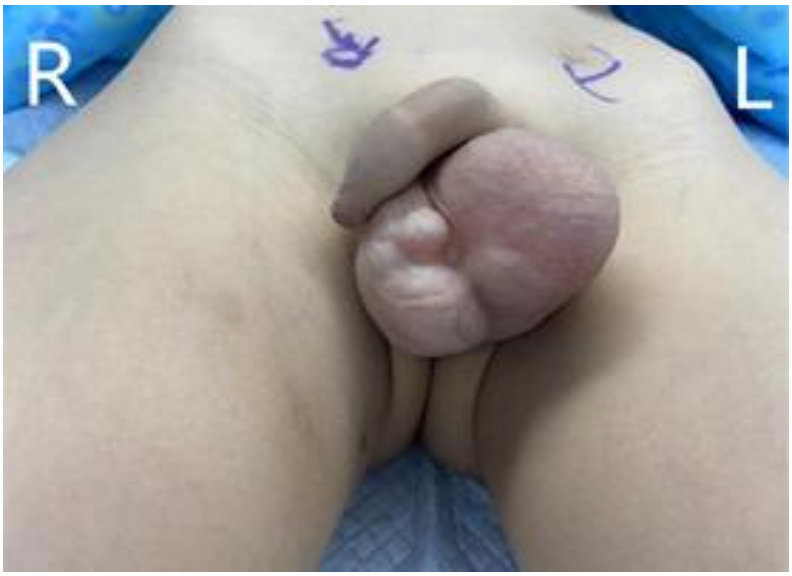


**Figure 1**: Bilateral scrotal masses for six months.

Denied history of diabetes, infection, or trauma. Six months ago, the parents found bilateral scrotal enlargement without obvious inducement, further ultrasound proved bilateral cystic masses in the scrotum, and lymphatic malformations were not excluded (Figure 2).

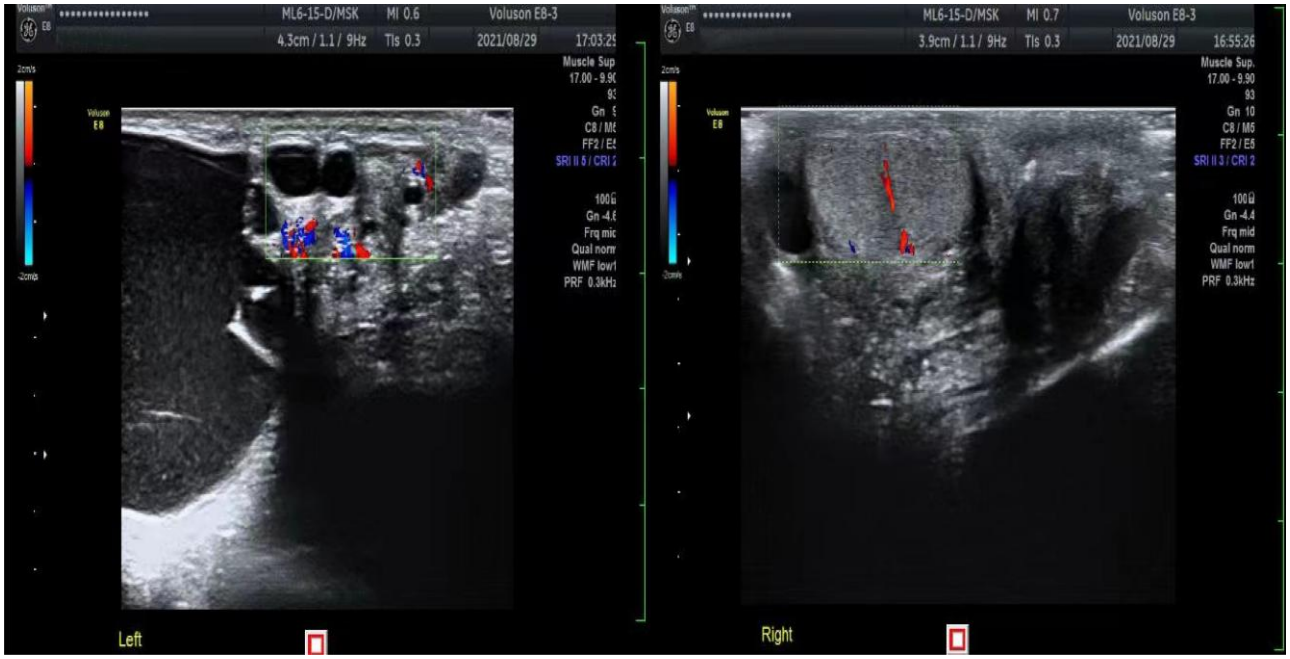


**Figure 2**: Bilateral cystic masses in the scrotum, and lymphatic malformations were not excluded.

Previously, "incarcerated hernia release + orchiopexy" was performed in the neonatal period due to "incarcerated hernia combined with transverse testicular ectopia". During the previous surgery, both testicles were found to be located in the left scrotum, and both testicles showed ischemic changes. One testis was hyperemic and with edema, and the other testis was completely black. No color change was observed after 10 minutes of warm saline hydropathic compress. No bleeding was observed after testicular white membrane incision. Thus, this testicle was assessed as necrotic. The parents were informed of the condition, and the remove

of the black testicle was highly recommended. But due to the parents' final decision, the black testicle was still being reserved. The patient recovered well after surgery. Regular ultrasound examination after discharge showed that both testicular volumes increased with good blood supply (Figure 3).


**Figure 3**: Regular ultrasound examination after discharge showed that both testicular volumes increased with good blood supply.

After this admission, combined with medical history, physical signs and ultrasound examination, Bilateral scrotal epididymal cyst or lymphangioma was considered. Lymphangioma is a benign lymphatic lesion which is normally caused by congenital malformation. Since scrotal lymphangioma can extend to perineum, inguen and even abdomen, it is easy to recur locally from the remaining lymphoid tissue [5]. Therefore, preoperative enhanced pelvic and abdominal MR was performed to accurately assess the location and extent of the lesion (Figure 4).

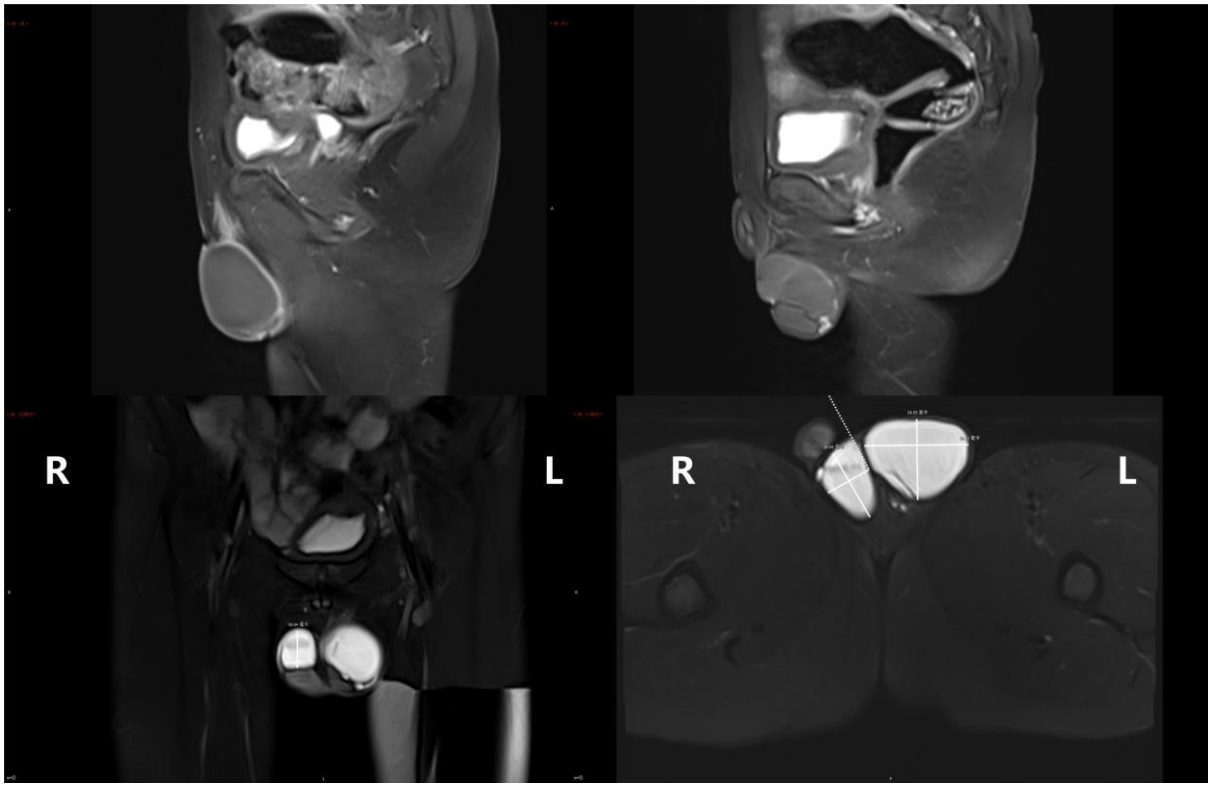

**Figure 4**: Preoperative enhanced pelvic and abdominal MR was performed to accurately assess the location and extent of the lesion.

The transverse incision of the right scrotum was taken. After cutting the skin, membrane and separate lymphatic tissue, surgical scar was seen and testicular sheath adhesion was severe. Testicle and epididymis were separated. The deferent duct was dilated. The lesion did not deviate to the inguen. The left and right lesions were connected by an intermediate tubular structure (Figure 5).

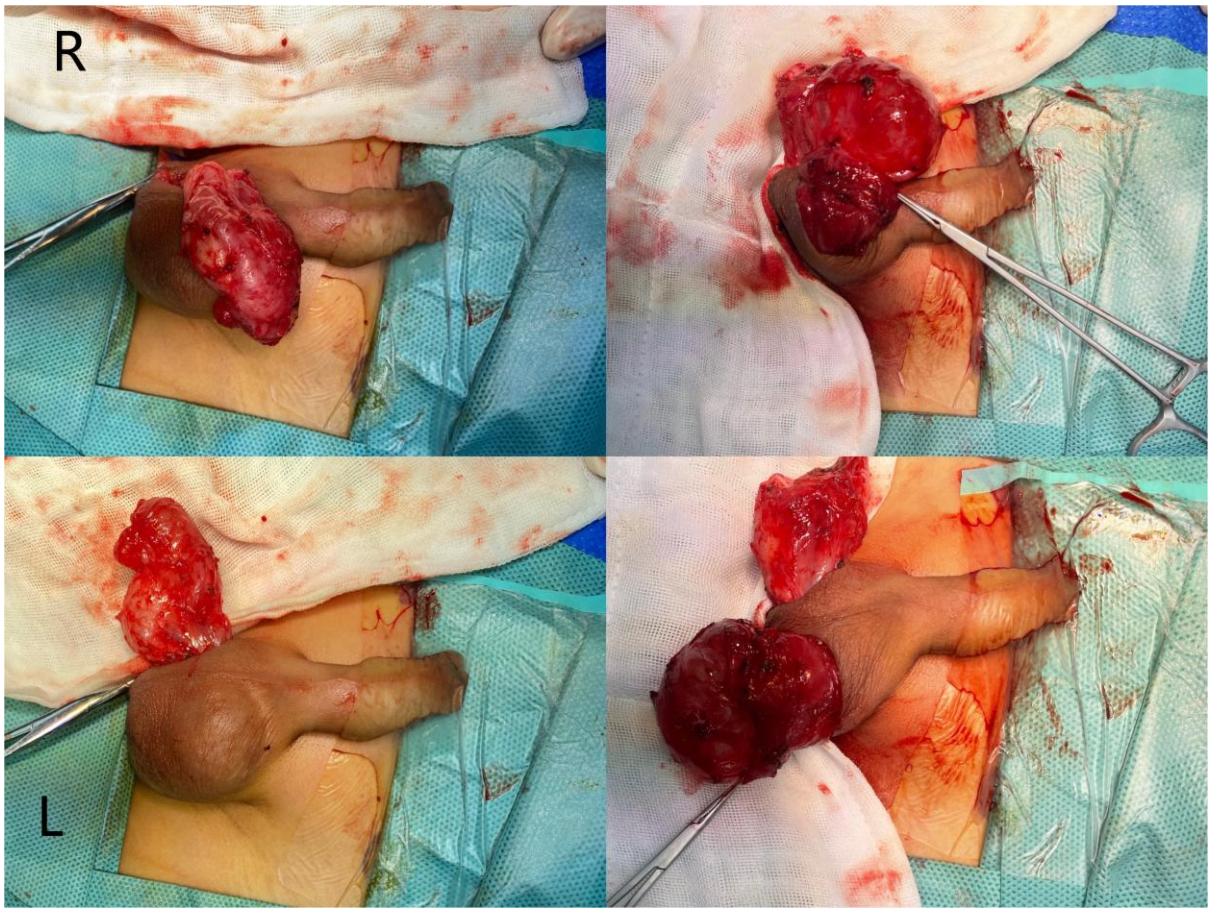


**Figure 5**: The left and right lesions were connected by an intermediate tubular structure.

The same procedure was performed on the left side. Complete resection of bilateral lesions and orchiopexy was performed. The operation was successful and the patient recovered well. Postoperative pathological diagnosis was: Bilateral scrotal epididymal cysts (Figure 6).

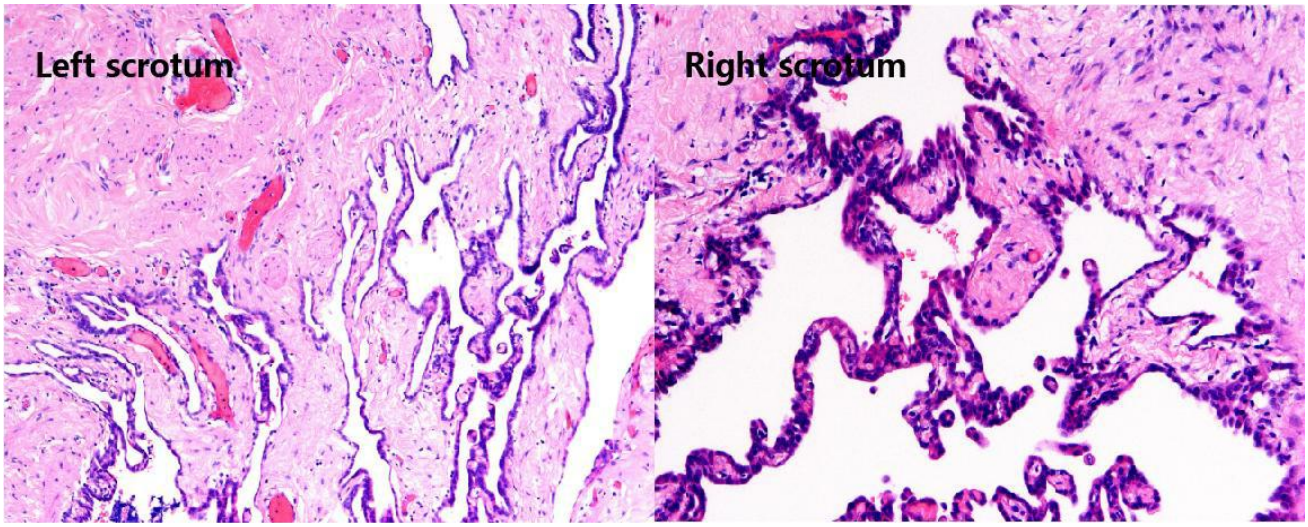


**Figure 6**: Postoperative pathological diagnosis was: Bilateral scrotal epididymal cysts.

The child was followed up for 2 months, and all index and activities were turned out normal.

## Discussion

Epididymal cyst is a benign fluid-filled sac in the testicles. Epididymal cyst can occurs anywhere within the epididymis (the narrow, tightly coiled tube above the testicle connecting the efferent ducts to the vas deferens) and does not contain sperm. Morbidity is estimated to be between 5-20% and increase with age [4,6-8]. There is no consensus on the pathogenesis of epididymal cyst. So far, various theories can be read in the literature. Major views consider epididymal cyst as an acquired disease, which usually presents as painless swelling of the scrotum. Usually caused by previous injury or inflammation. Epididymal cysts are collection of fluid in a single sac (unilocular) or more than one (multilocular) as a result of dilatation of efferent epididymal tubules due to tubular obstruction. A study found that local production of proinflammatory cytokines evidenced by elevated concentrations of interleukin 8 (IL-8), interleukin 6 (IL-6) in epididymal cyst is responsible for cyst formation. Most are unilateral, and bilateral epididymal cysts are rare [4,9,10]. Some other authors suggest that this may be related to testicular dysplasia syndrome. The incidence of epididymal cysts increases during adolescence. The syndrome may be caused by various endocrine disruptors during embryonic growth and development [8,11]; Other authors suggest that epididymal cyst is a new congenital lesion of the epididymis, called congenital cystic dysplasia, and might be associated with renal and/or urinary tract malformations [12]. In this case, due to incarcerated hernia and transverse testicular ectopia during the neonatal period, hernia contents prolonged and compressed spermatic cord structure, resulting in scrotal injury and testicular ischemia. Seven years later, the patient developed bilateral scrotal epididymal cysts. This may be associated with previous inflammation and injury caused by epididymal efferent tubules compression from the transverse testicular ectopia and incarcerated hernia. In this case, transverse testicular ectopia and incarcerated hernia occurred simultaneously during the neonatal period, and the sac contents prolonged and compressed spermatic cord structure, causing one of the testis' ischemia, necrosis. And the other one's hyperemic, edema [13]. Infants younger than 6 months are lack of abundant collateral vascular structure. Thus, are more likely to cause testicular ischemia due to incarceration [14]. Testicular necrosis refers to the stop of metabolism and loss of function of the testicular tissue cells due to the obstruction of blood circulation. The clinical feature would be testicular atrophy. During the operation, the surgeon used lukewarm saline to compress the testis for 10 minutes and also cut the testis tunica vaginails and showed no signs of blood reperfusion [15]. Thus that testicle was judged to have complete ischemic necrosis. In order to avoid the necrotic testis causing inflammation or cancerate. The full resect of the necrotic testis was recommended. But the family strongly requested that both testicles be preserved, thus the surgeons retained and immobilized both testicles. To our surprise, during the follow up examinations. Nine Inguinal and scrotal ultrasounds presented the "necrotic" testicle showed volume growth and well-distributed echo. During the bilateral scrotal mass resection of the second hospitalization. We also observed that the "necrotic" testicle had normal blood flow and function except that it was smaller than the other side. Mustafa Yasar Ozdamar [16] pointed out in the literature that testicular ischemic necrosis caused by incarcerated oblique inguinal hernia in infants can be treated conservatively without removing the testicle, and ortopexy can be performed after wet compress with lukewarm saline for 10 minutes. After surgery, the children were followed up for 6 months, and the testicular size and arterial blood flow were evaluated by color Doppler ultrasonography. In his study, 44 incarcerated inguinal hernias resulted 9 (20.4%) testicular ischemic necrosis of varying degrees. During 6 months' follow-up, testicular atrophy occurred in only 2 patients (22.2%). The testicular volume was reduced by 10%. Jonathan D. Kaye [17] pointed out in his report that 37 cases of testicular ischemic necrosis were caused by testicular torsion, and 34 cases of them were performed testis resection which was confirmed as necrotic by postoperative pathology. 3 cases retained the necrotic testis according to the parents' wishes, and testicular atrophy were found in all 3 cases during postoperative follow-up. However, all cases were older than one-year-old and were not neonates. There are few reports on testicular necrosis in neonates. In addition, there is still no unified standard for testicular inactivation time. Therefore, the possibility of intraoperative misdiagnosis cannot be ruled out. Pietr. H. Callewaert [18] pointed out in the literature that whenever

possible, for bilateral testicular ischemic necrosis caused by neonatal scrotal emergency, necrotic testicle should be preserved as much as possible, for some testicular functions can still be realized [15]. In this case, a total of 10 inguinal and scrotal ultrasound examinations were performed before and after the first surgery. Lambert's empirical formula was used to calculate testicular volume: Volume = Length (L)× width (W)× height (H)×0.71 [19]. There was differential testicular ischemia on both sides (more severe on the left side) due to incarcerated hernia and transectopic testicle. After surgery, the volume of bilateral testis increased, the echo was even, and blood supply was restored. In our case, the testicles were inactivated due to the scrotal emergency. But postoperative volume can still increase and blood flow recovery can still occur. It may be related to the strong testis ischemia tolerance in neonates. This case report is helpful to determine whether to choose orchiectomy or conservative orchiopexy to avoid testicular resection for testicular ischemic necrosis caused by the scrotal emergency of neonates. Lymphangioma is a benign lymphatic lesion. It is commonly believed that lymphangioma is caused by congenital malformation of lymphatic tissue. However, it can also occur after local injury of lymphatic vessels, like lymphatic infection, inflammation, trauma, Surgery or radiation etc. This is called acquired lymphangioma. It is very rare to see acquired lymphangioma involving the scrotum. A detailed literature review in English on PubMed found only 12 cases of acquired lymphangioma involving the scrotum [6,20]. The main manifestation of Scrotal lymphangioma is scrotal distension. Typically described as round, translucent or fuzzy groups of variously sized vesicles resembling frog eggs [21]. Ultrasonography of scrotal lymphangioma shows multiple thin septa and compartments in both scrotums. No internal vascular structure is shown. Lesions may extend along the scrotal wall, the groin area, and the perineum [5]. Epididymal cyst ultrasonography shows a thin wall separating cyst in epididymal head with echo [8]. Both of them showed similar ultrasound performance, showing water signal, cystic change, clear boundary, no fat signal, no calcified strong echo mass. They are also similar in megascopic structure, with multilocular cystic structure, smooth cyst cavity, and the liquid in the cyst cavity is mainly light yellow and clear liquid. lymphangioma is consist of dilated lymphatic vessels with irregular lumen under microscope. Dilated lymphatic vessels contain protein fluid. The inner lining of the dilated lymphatic vessels is lined with flat and widely spaced endothelial cells. There is an overgrowth of adipose tissue and smooth muscle hyperplasia Is not obvious [5] The wall of epididymal cyst is composed of fibrous tissue. Lined with a single cubic epithelium. It is consists of smooth columnar epithelium with an inner layer containing serous fluid. Epididymal cyst is mainly unilateral, bilateral simultaneous occurrence is rare. Lymphangioma of the scrotum is commonly reported in individual cases and may extend to the perineum, groin and even abdomen. Prone to local recurrence from residual lymphoid tissue. Both are benign lesions, The clinical manifestations of them were similar. The preferred adjunctive examination is ultrasound. The most effective treatment is complete surgical excision of the lesion. MRI can confirm the diagnosis and lesion extent before surgical resection. The final diagnosis depends on postoperative pathological findings.

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## Authors' Contributions

MKS and HW contributed to study conception and design. JLH collected and analyzed clinicopathological data and wrote the manuscript. GFL was involved in data analysis and article translation. SRS and JGW SLL analyzed the article and provided suggestions. The final version of manuscript was read and approved by all authors.

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## Consent for Publication

The patient consented to publish this case report and associated images.

## Competing Interests

None of the authors declares a conflict of interests.

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