

Inflammation and infection

## Rare cause of acute scrotal swelling and pain in adult- a case report on spontaneous pampiniform plexus thrombosis and literature review

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### ABSTRACT

Acute scrotal pain is a common urological emergency where clinical judgement plays pivotal role in making the diagnosis. Scrotal pain secondary to spontaneous pampiniform plexus thrombosis is rare, and we are the first who report in the Asian region.

A 50-year-old gentleman presented with left-sided scrotal pain & swelling. The diagnosis of pampiniform plexus thrombosis was confirmed by the ultrasound Doppler. He was treated with anticoagulant and good clinical result was obtained.

Uncomplicated thrombosis could be managed non-surgically with good clinical outcome and radiological resolution. Ultrasound doppler remains first-line investigation to establish diagnosis, monitor the progression and treatment outcome.

### Introduction

Acute scrotal pain is a common urological emergency which has a vast array of differential diagnoses ranging from the usual conditions such as epididymo-orchitis and testicular torsion to the infrequent conditions like hematocele, infected hydrocele, pyocele and necrotic testicular malignancy. Testicular torsion should always be ruled out in the event of young patients until proven otherwise. Scrotal swelling or pain secondary to spontaneous pampiniform plexus thrombosis is rare, and so far, there have only been 20 cases reported in the literature. Spontaneous thrombosis of pampiniform plexus was initially described by Senn (1902 & 1904) and then reported by McGavin in 1935.<sup>1</sup> This case would represent the first reported case in the Asian region.

### Case presentation

We are reporting a middle-aged man, who presented with left-sided scrotal pain and swelling for one week. The pain had begun insidiously and increased in intensity gradually. He had no fever or vomiting. He also denied any urinary or bowel symptoms. There was no similar episode previously, and he denied any trauma to the scrotal region. His past medical and surgical history were unremarkable. He did not take

any medication, illicit drugs, nor traditional herbs.

On examination, the patient was afebrile. Bilateral testes were palpated, and they were in normal lie presentation. The left epididymis appeared to be mildly swollen; however, the overlying skin was not erythematous. Minimal tenderness was appreciated at the patient's left hemi-scrotum, along with the left spermatic cord. The remaining physical examination was unremarkable. His full blood count showed mild leukocytosis and coagulation profile was normal. His urinalysis only showed trace leucocytes with negative urine cultures.

The ultrasound Doppler imaging of testis and scrotum was performed by our radiologist on the same day of consultation. Both testes had homogenous parenchymal echotexture with good vascularity and no abnormal focal lesions within. The left testis was 4.45 cm (CC) x 1.73 cm (AP) x 2.72 cm (width) whereas the right testis was 3.92 cm (CC) x 1.90 cm (AP) x 2.77 cm (width). The left scrotal sac was thickened and edematous (Fig. 1). There was engorged, dilated slow flow of pampiniform plexus over the left scrotum; thus, radiological report concluded the diagnosis of left-sided pampiniform plexus thrombosis (Fig. 2). The ultrasound of the rest abdomen was normal. The patient was admitted into urology ward, advised for bed rest and treated expectantly. He was prescribed with low molecular weight anticoagulant enoxaparin sodium 60 mg given twice a day subcutaneously. He was also given intravenous

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antibiotic Amoxicillin-Clavulanate to cover for epididymitis and oral analgesia for pain control. His pain and swelling subsequently subsided. The repeat ultrasound was carried out six days later and it showed complete resolution of pampiniform plexus thrombosis (Fig. 3). This patient was planned for the continuation of oral anticoagulant (tablet warfarin 3mg daily) for a total duration of one month. No surgical exploration was required, and the patient was well upon review.

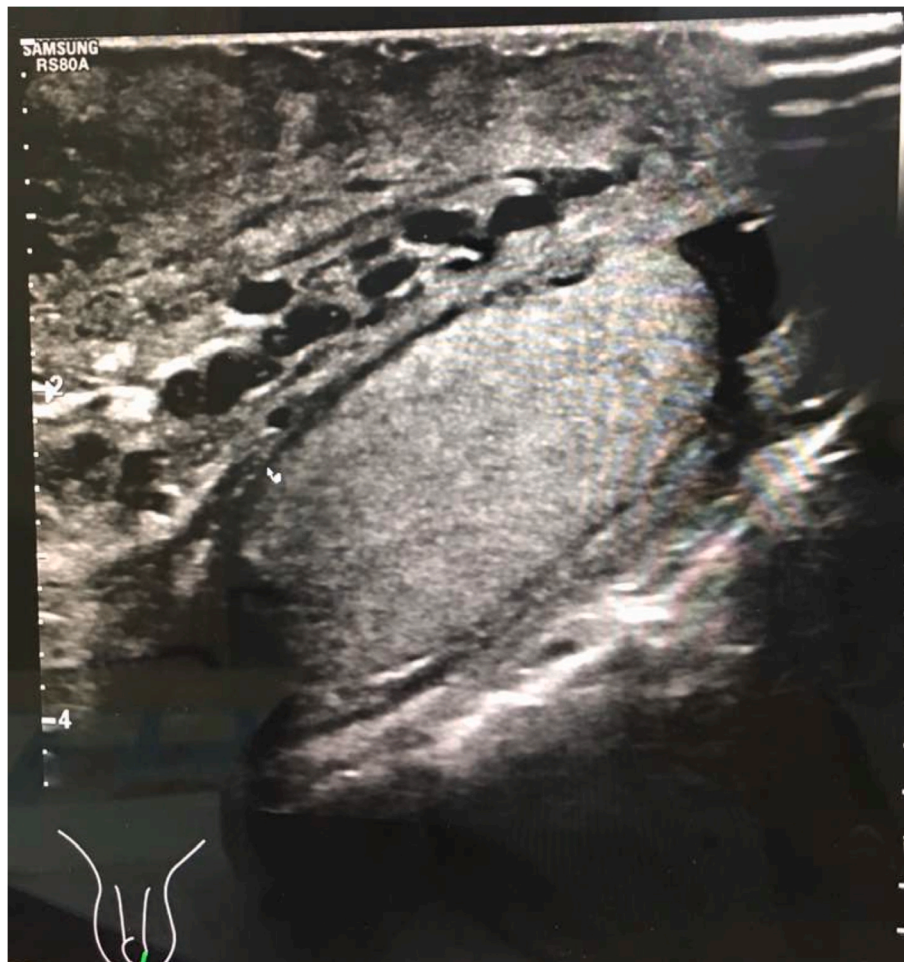
## Discussion

Diagnosis sometimes can be challenging in case of scrotal swelling with pain. It can be misdiagnosed as testicular torsion, orchitis or even incarcerated inguinal hernia, which in turn resulted in unnecessary surgical intervention.<sup>2,3</sup> Spontaneous pampiniform plexus thrombosis is a very rare presentation. Left sided thrombosis is more common compared to the right based on the cumulative case reports. The exact pathophysiology is unknown, however, theoretically anatomical factors for the preponderance of varicocele of left side might explain the prevalence of left side occurrence. The left gonadal vein drains into left renal vein perpendicularly rendering it at a higher risk of more significant pressure but lower blood flow.<sup>4</sup> Vicrow's triad is responsible for the pathology of thrombus formation and raised intraabdominal pressure might reduce the gonadal vein blood flow; therefore, creating an environment of stasis and thrombosis. Other etiologies such as retroperitoneal tumours, incompetent valves, compression of renal vein by superior mesenteric arteries were among the rare causes of thrombosis.<sup>4,5</sup> Medical condition like hypercoagulable state ought to be considered as well.

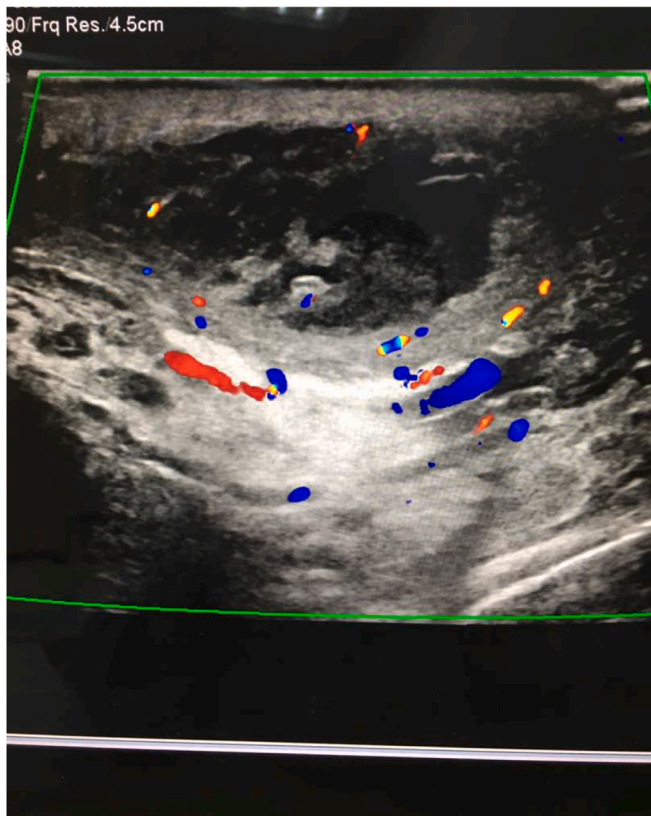
However, in our case, we could not determine any cause or risk factor for this patient.

The commonest preoperative diagnosis was a complicated inguinal hernia among all the reported cases, and most of them had undergone surgical exploration. More than half of the patients went through surgical excision. In 1985, Roach et al. reported that the patient who was treated with excision had resulted in orchidectomy due to the complications of venous congestion and hematoma. Therefore, in this context, we believe that conservative treatment may be considered as a better management strategy.

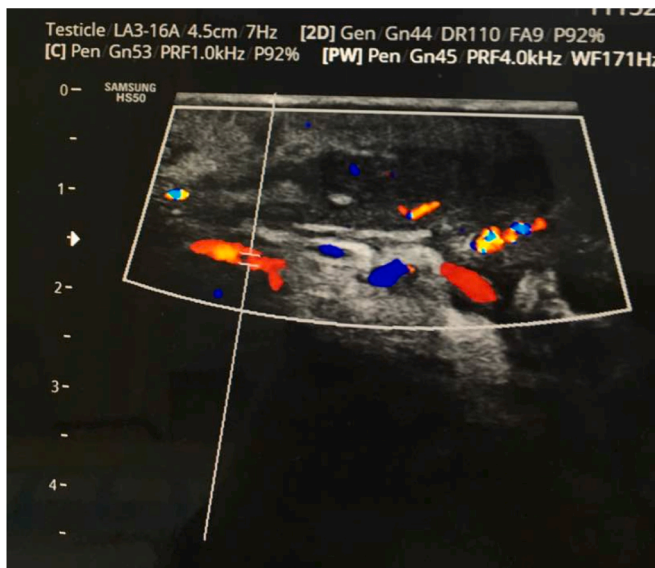
Due to restricted references in the literature, there are no guidelines for the management of pampiniform plexus thrombosis. It is paramount important to determine the causes of the thrombosis. Several investigations have been outlined in the previous case reports include doppler ultrasonography, detailed CT imaging, blood screening or even surgical exploration to establish the causes. However, examination with ultrasound doppler should be the first line as it is a non-invasive and sensitive means to establish the diagnosis. In this case, ultrasound doppler helps to provide us with a clear diagnosis. Once the diagnosis is confirmed, anticoagulant should be prescribed to treat thrombosis unless contraindicated. Repeat imaging with ultrasound doppler is adequate to monitor the progress of disease and treatment outcome. In our opinion, MRI or even computed tomogram is not necessarily done due to the risk of radiation, extra cost, with no added benefits. There is no role to excise the thrombosed plexus, as evidenced by the good results in our case and at least three other cases.



**Fig. 1.** Sagittal view ultrasound of left testis showed normal homogenous parenchymal echotexture. There was no obvious focal lesion seen with the testicular parenchyma. However, the left scrotal sac was edematous and thickened.



**Fig. 2.** Ultrasound Doppler of the scrotum revealed engorged, dilated and slow flow of pampiniform plexus of left testis. There are some areas of none or sluggish flow suggestive of thrombosis. There was a focal dilated vein in the most inferior part of left scrotal sac.



**Fig. 3.** Ultrasound performed on day 6 demonstrated less echogenicity in the previously engorged and dilated pampiniform plexus. It showed good doppler signal and its features was consistent with a resolution of left pampiniform plexus thrombosis.

## Conclusion

Pampiniform plexus thrombosis is indeed an uncommon clinical

entity in urology where the management remains controversial. The pathophysiology is not very well understood. Good history taking, proper examination and precise investigations are required to confirm the diagnosis of acute scrotal pain. Ultrasound Doppler remains the first line investigations to establish the diagnosis of thrombosis. In this clinical case study, spontaneous pampiniform plexus thrombosis may be safely and effectively managed conservatively with the anticoagulant.

## Author contributions

The first author contributed to this paper in terms of acquisition and analysis of literature, drafting the manuscript and critical revision. The other authors are mentoring and appraising as well as proofreading the manuscript.

## Conflicts of interest

The report is self-funded. We declare no conflicts of interest.

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