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Perinatal Torsion of Appendix Testis

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1. Abstract

Torsion of the appendix testis is an extremely rare cause of scrotal swelling in neonates. We report two cases of torsion of the appendix testis in a one-day-old male and a one-month-old male. Their clinical and radiological features are discussed. Torsion of the appendix testes should be included in the differential diagnosis of scrotal swelling in newborns and nonoperative therapy is recommended to prevent surgical exploration and its associated risks.

2. Introduction

Testicular torsion results from twisting of the spermatic cord within the tunica vaginalis, followed by venous congestion and if not recognized and treated promptly leads to loss of arterial blood supply, and subsequent ischemia and necrosis of the affected testis. Approximately 86% of testicular torsions occur in males older than 10 years (median age, 15 years) [1]. This is called intravaginal testicular torsion to differentiate it from the perinatal testicular torsion which is commonly extravaginally.

Testicular torsion that occurs in the first month of life is referred to as perinatal testicular torsion. This is further subclassified as prenatal or intrauterine and postnatal or extrauterine torsion. Intrauterine and extrauterine torsion differ in terms of their timing, clinical presentation, and salvage rates. While intrauterine testicular torsion presents immediately after birth often with painless, swollen and discoloured scrotum, extrauterine testicular torsion occurs later within the first month of life and presents with acute scrotal pain, swelling and redness [2-4].

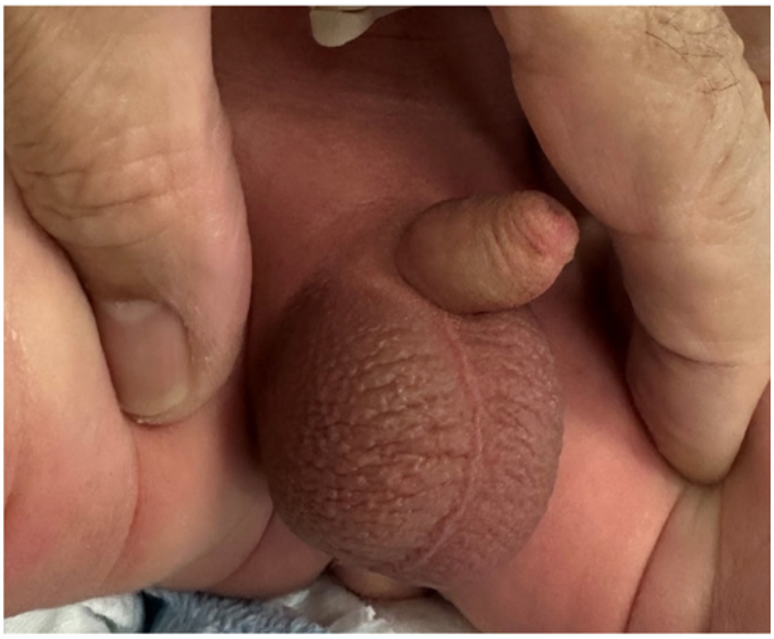
Torsion of testicular appendages is considered one of the most common causes of acute scrotal pain and swelling in prepubertal children. Therefore, it should be included in the differential diagnosis for any male presenting with an acute scrotum. There are several causes of scrotal swelling in the neonate. The most common causes include intrauterine testicular torsion, neoplasms, supernumerary testis, splenogonadal fusion, and adrenal rests [5,6]. Torsion of the appendix testis on the other hand is an extremely rare cause of scrotal swelling in the neonatal period [7,8]. We present two cases of torsion of the appendix testis in a newborn male child and a one-month-old male child. Aspects of diagnosis and management are discussed, and the literature is also reviewed.

3. Case Reports

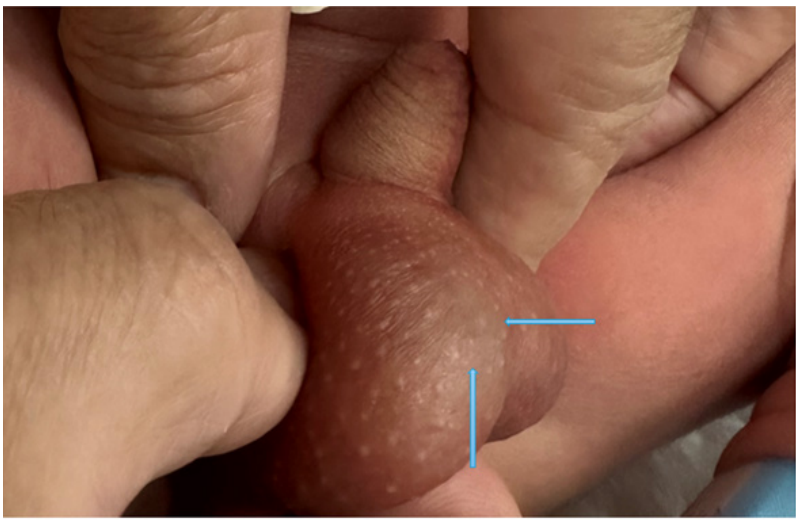
3.1. Case No 1

A one-day male newborn was noticed to have right scrotal swelling. He was a product of normal spontaneous vaginal delivery. His birth weight was 3.2 kg. Clinically, he was found to be healthy with no other abnormalities. He passed meconium normally. He was found to have right hydrocele which was not tender but slightly tense. The hydrocele showed what looks like the blue dot sign which represents the swollen appendix testis within the scrotal sac that has a blueish colour because of loss of its blood supply (Figures 1A and 1B).

He started on oral feeds, and he tolerated oral feeds well. Scrotal ultrasound revealed both testes are present in the expected location and of normal size. There is no intratesticular mass on either side, with normal homogenous appearance of both testes. The left testicular appendix was noted and appeared normal (Figure 2). The epididymis was normal bilaterally. On the right side there was a mild vaginal hydrocele of turbid fluid. There was a floating well-defined rounded-shaped avascular, freely mobile lesion which measures about 7mm in diameter of heterogenous echo pattern (echogenic outline-central hypo-echogenicity with cystic changes and interlacing fibres) (Figures 3, 4, 5, 6 and 7). The diagnosis of right mild turbid infantile hydrocele with floating avascular lesion suggestive of detached testicular appendix with necrotic changes. He was treated conservatively and on follow-up 8 weeks later he was doing well and a repeat scrotal ultrasound showed disappearance of the right hydrocele and the floating avascular mass.



Figures 1A and 1B: Clinical photographs showing a newborn with a right hydrocele. Note what looks like the blue dot sign seen in the second photograph representing the detached appendix testis.



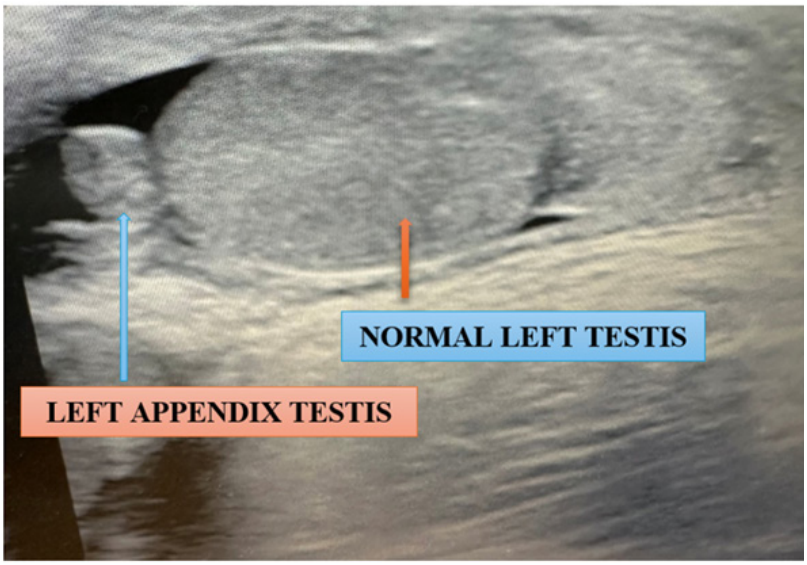
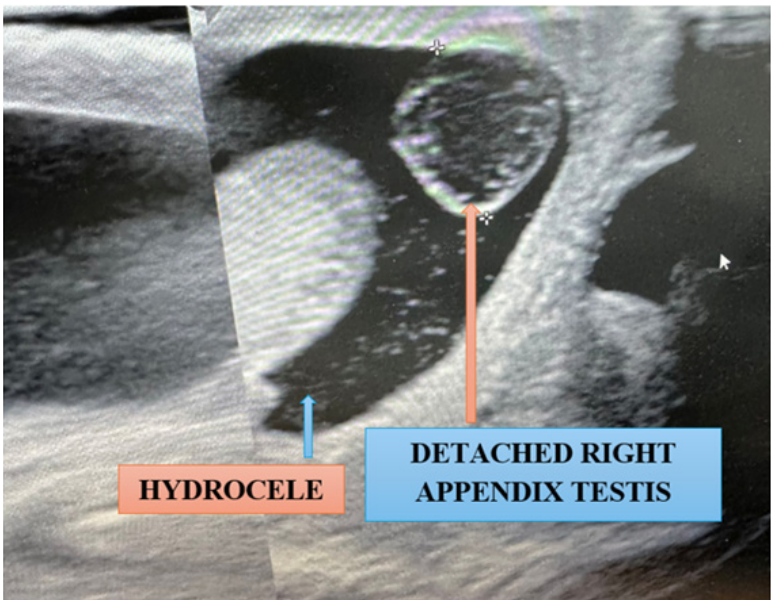


Figure 2: Ultrasound of the scrotum showing a normal left testis and a normal left appendix testis.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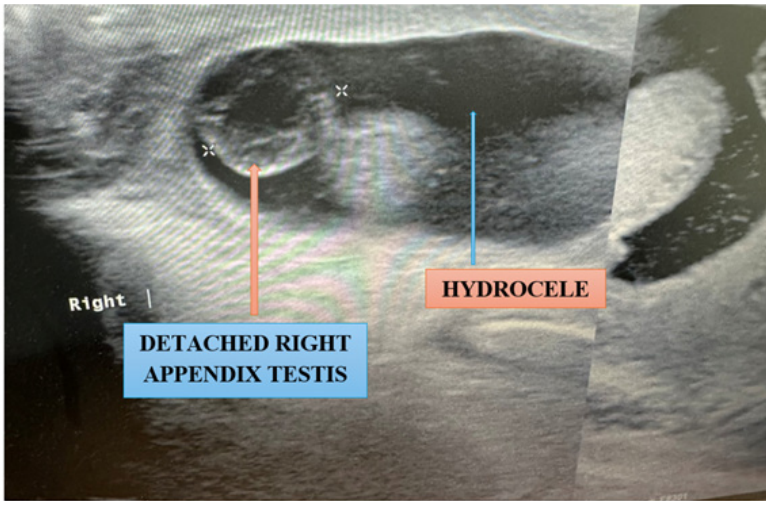
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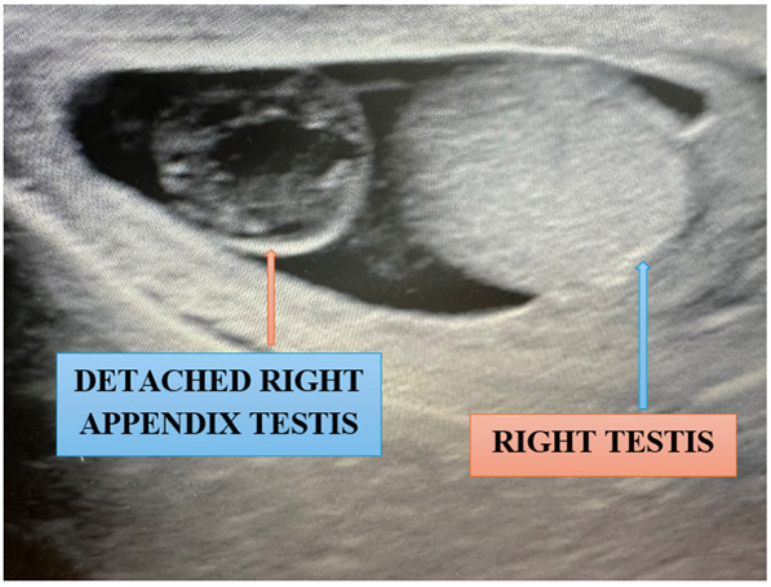
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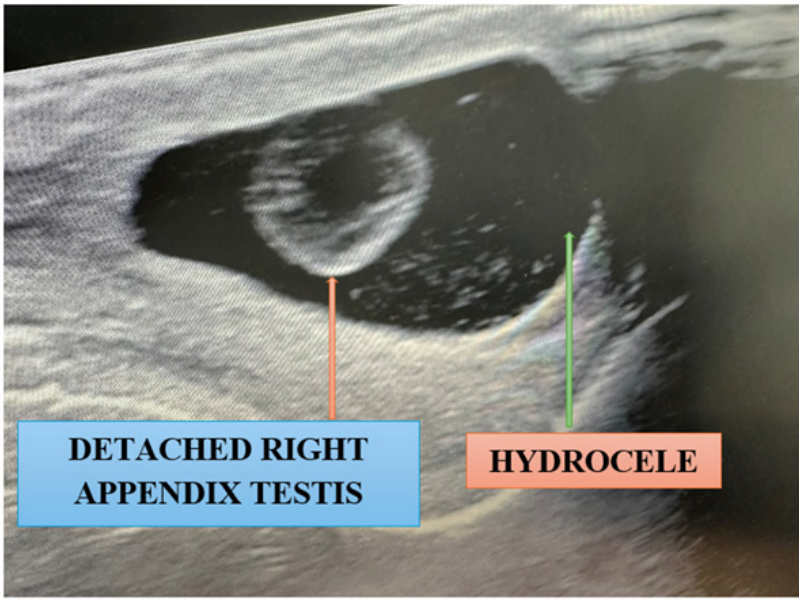

Figures 3, 4, 5 and 6: Ultrasound of the scrotum showing the detached right appendix testis. Note the associated hydrocele and the detached appendix testis moving in different positions within the hydrocele.

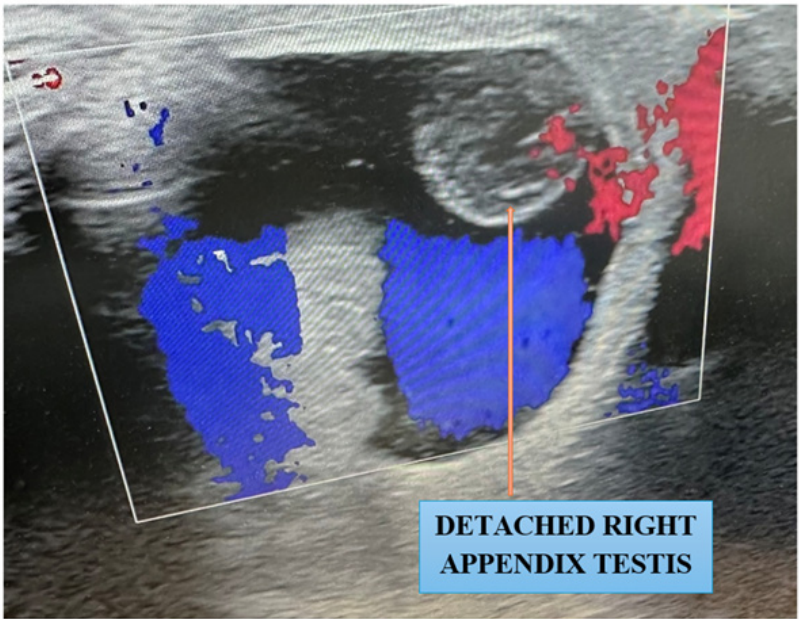

Figure 7: A color Doppler Ultrasound of the scrotum showing the detached right appendix testis which appears avascular with no blood supply.

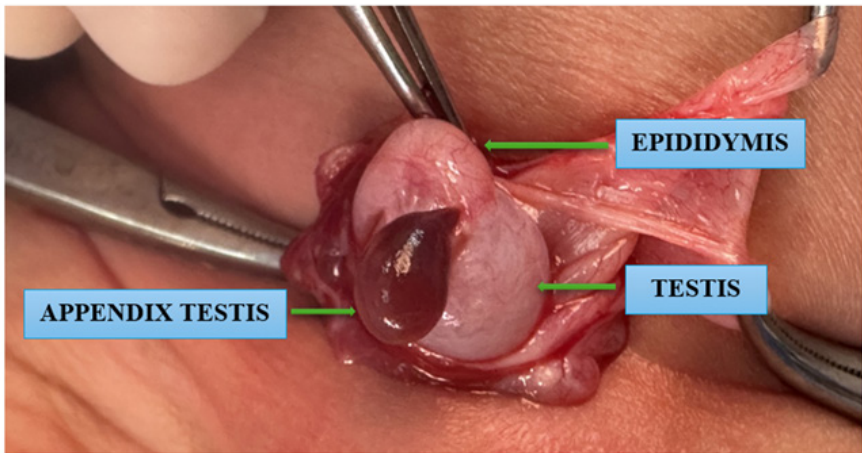

Figure 8: A clinical intraoperative photograph showing torsion of the appendix testis. Note the darkish discoloration of the appendix testis because of ischemia.

3.2. Case No 2

A one-month-old male was referred to our hospital with right inguinal swelling.

He was a product of normal spontaneous vaginal delivery. His birth weight was 3.8 kg, and his clinical examination revealed no abnormalities. At the age of one month, he started to develop a right inguinal swelling. The swelling appeared one week prior to presentation to our hospital and increased gradually but still was reducible. Clinically, he was found to have a large inguinal hernia. The hernial sac contained intestines that were reducible. Ultrasound confirmed the diagnosis of right inguinal hernia and no other pathology. He underwent right inguinal herniotomy and during exploration, he was found to have torsion of the testicular appendix that was blackish in colour (Figure 8). This was excised and right inguinal herniotomy was done. Postoperatively, he did well and was discharged home on the same day. He was seen one week later in the clinic and was well.

4. Discussion

There are two important embryological remnants related to the testes, the appendix testis and appendix epididymis (9). The appendix testis, which is also called hydatid of Morgagni, is a vestigial remnant of the Mullerian duct and is present in 76% to 83% of testes. The morphology of the appendix testis is variable and can vary from a small nodule to a longer protuberance. When present, it is located on the superior pole of the testicle between the testis and epididymis. It is homologous to the female's fimbriated end of the Fallopian tube. The appendix testis is the most common testicular appendage to undergo torsion. The appendix epididymis is a vestigial remnant of the Wolffian (mesonephric) duct present in 22% to 28% of testes. When present, it occurs along the head of the epididymis. Torsion of these remnants can occur in longer pedunculated appendices, compromising their blood supply. Torsion of testicular appendages is considered one of the most common causes of acute scrotal pain in prepubertal children. Torsion of a testicular appendage is most commonly observed in boys between the ages of 7 and 12, although it can occur at any age. It is important to note that more than 50% of boys presenting with acute scrotal pain are diagnosed with torsion of a testicular appendage [14-16].

The appendix testis was first described by Morgagni in 1761. Embryologically, it represents persistence of the upper end of the mullerian duct in males. Although it can present as a sessile swelling but more frequently seen as a pedunculated structure arising from the superior pole of the testicle adjacent to the head of the epididymis and its size is variable ranging from 1 to 7 millimetres long. Torsion of the appendix testis is the most common cause of an acute painful microtome in a child. It is commonly seen in boys aged 7–14-year-old. The usual presentation is with acute scrotal pain and swelling in the anterosuperior region of the testicle [5,6,10,13]. There is typically a pathognomonic "blue dot" sign, which represents the swollen appendix testis within the scrotal sac that has a blueish colour. The tossed appendix testis may be palpated as a 2-3 mm firm nodule in the upper pole of the testicle. These patients usually do not experience systemic signs of fever, nausea, or vomiting. Most patients with torsion of the appendix testis and once the diagnosis is confirmed can be treated conservatively with pain control and rest. Occasionally the pain persists or if the diagnosis cannot be ascertained, surgery may be required to confirm the diagnosis and expedite the recovery process.

Torsion of the appendix testis in the neonatal period on the other hand is exceedingly rare. It is a rare cause of scrotal swelling in the neonatal period. There are several causes of scrotal swelling in the neonate. The most common cause is hydrocele. Other causes include intrauterine testicular torsion, neoplasms, supernumerary testis, splenogonadal fusion, and adrenal rests [5,6]. One of the most important causes of scrotal swelling in newborns is intrauterine testicular torsion. Torsion of the testis in the newborn usually presents as an enlarged, firm, nontender, nontrans illuminating mass in the scrotum without signs of systemic reaction or toxicity. It is usually fixed to the overlying skin which is discoloured. This condition is known to occur in full-term babies who are of above-average birth weight, and it is usually unilateral, but simultaneous bilateral cases have been reported [14, 15,16].

In 1969, Chiles and Foster Jr described the first case of torsion of the appendix testis in a newborn [7]. The diagnosis was confirmed after surgical exploration at the age of 18 hours. Their patient was originally thought to have testicular torsion and treated with scrotal exploration. At surgery it was discovered that the patient had torsion of the appendix testis and the cord itself was normal. Krishnan et al in 2016 reported a newborn with torsion of the appendix testis [8]. Their patient was diagnosed clinically and radiologically and was treated conservatively. This is similar to our patient who had an extra testicular mass and a hydrocele. Our patient was treated conservatively and on follow up, the extra testicular mass and the hydrocele disappeared. Our second patient was diagnosed as an incidental finding during right inguinal herniotomy.

In conclusion, torsion of the appendix testis is an extremely rare cause of scrotal swelling in neonates. Torsion of the appendix testis should be included in the differential diagnosis of the causes of scrotal swelling in the neonate. Ultrasonography is an important investigation in the diagnosis of a neonate with scrotal swelling. Physicians caring for these patients should be aware of this and if torsion of appendix testis is suspected and other more serious causes of scrotal swelling such as testicular torsion and irreducible inguinal hernia are excluded, conservative management can be considered.

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