CASE REPORT

Twisted intra-abdominal cyst in a neonate: a surprise revelation

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SUMMARY

We, herein, present a male neonate with an antenatally detected intra-abdominal cyst who presented at 18 days of life at which time, the ultrasound revealed a 5×4 cm cyst. Since he was asymptomatic, we planned to repeat the ultrasound a month later and operate if the cyst showed no regression. However, a week later, he presented with an acute abdomen, irritable cry and a repeat ultrasound showing a larger (8×6 cm) cystic mass with debris within. He was taken up for an emergency laparotomy. Intraoperatively, the cyst was found arising from the left lateral abdominal wall free from all structures with a twisted pedicle. Histopathology surprisingly revealed seminiferous tubules within the cyst wall with the vas deferens, thus confirming the diagnosis of a torsion of intra-abdominal testis. Hence, we emphasise the importance of examining for an undescended testis when dealing with a male neonate presenting with a cystic intra-abdominal mass.

clinically described in older children and has varied clinical presentations leading to delayed diagnosis. This condition, however, presenting as a cystic neonatal abdominal mass is extremely rare and torsion of this cystic mass presenting with an acute abdomen is rarer still. Less than 40 cases of torsion of intra-abdominal testes have been reported worldwide as cited in this journal in 1997.⁴ This is the first case being reported in the Indian subcontinent.⁵ This is the third reported case of a neonate with a torsion of an undescended testis presenting as an intra-abdominal cyst the world over, to the best of our knowledge.^{6,7}

The incidence of testicular torsion in undescended testis is much higher than a completely descended one for reasons unknown.⁸ Torsion of an undescended testis can be easily picked up on clinical examination. When considering the differential diagnosis of such a large cyst, testicular origin is hardly ever thought of.⁹

BACKGROUND

The diagnosis of an intra-abdominal cyst in a fetus poses a diagnostic dilemma because of the large number of potential differential diagnoses to be considered. Prediction of the neonatal course is impossible as it varies from complete resolution to life-threatening complications.¹ Common cysts include those arising from the gastrointestinal tract and the genitourinary tract.² Cysts arising from the testis is a difficult diagnosis to make both prenatally and postnatally.

Undescended testis is one of the most common paediatric condition for which surgery is performed.³ It affects 3% of all term babies and up to half of all preterm babies.³ Intra-abdominal torsion has been

CASE PRESENTATION

An 18-day-old male neonate was antenatally detected to have an abdominal cyst elsewhere and advised to follow up post delivery. Postnatal scan revealed an abdominal cyst of size 5×4 cm. As the child was healthy and feeding well, it was decided to put him on close follow-up with a repeat ultrasound after a month. However, they presented a week later with worsening abdominal distension and irritable cry for a duration of 1 day.

On examination, he was found to have a cystic tender, mobile mass of size 5×5 cm, which was found to occupy the left lower quadrant. All the margins of the swelling were felt.

The scrotal examination was unfortunately forgotten as the discovery of the abdominal cyst distracted us from a thorough clinical examination.

INVESTIGATIONS

His blood investigations revealed a total count of 22 200 with 56% neutrophilia. An ultrasound done the same day revealed a large cystic mass, 7.4×8.2 cm with 1–2 internal septations and debris seen (figure 1). Organ of origin could not be made out on imaging. At this point, a diagnosis of a possible lymphatic cyst with haemorrhage had been made and it was decided to take the patient up for an emergency laparotomy. He was administered intravenous antibiotics which included both Gram-negative and Gram-positive coverage and intravenous fluids preoperatively

Figure 1 Ultrasound abdomen showing the large cystic mass with internal septa.

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Figure 2 Intraoperative image.

TREATMENT

Intraoperatively, he was found to have an 8×8 cm torted, haemorrhagic cyst attached to the left lateral abdominal wall with no attachment to any organ whatsoever. It was not possible to make out an organ of origin so the diagnosis was in doubt. The twisted pedicle was cut between two haemostats and the pedicle tied with a silk ligature (figure 2). The rest of the abdomen was inspected and found to be normal.

Figure 3 Histopathology showing seminiferous tubules in the cyst wall.

Figure 4 Histopathology showing vas in the cyst wall.

OUTCOME AND FOLLOW-UP

He was administered intravenous antibiotics postoperatively for 24 hours after which they were stopped. Feeds were started on the second postoperative day, which was initially expressed breast milk in incremental amounts graduating to demand breast feeds which he tolerated.

He was discharged 4 days later and at discharge he was active and tolerating feeds well.

He was reviewed a week later at the outpatient department and found to be feeding well.

The histopathological examination revealed a surprise with seminiferous tubules (figure 3) and vas deferens (figure 4) seen in a bed of haemorrhage accompanied by haemorrhagic vessels within the cyst wall concluding with a diagnosis of a torted undescended intra-abdominal testis.

He was not offered prophylactic orchidopexy on the right side but was educated about the warning signs and the need to report immediately to the emergency ward if they were to notice any of those signs.

He was followed up over the course of a month and was doing well.

DISCUSSION

Fetal abdominal cysts are quite easily picked up on ultrasound imaging. However, the origin of these cysts is quite an arduous task to diagnose. The vast majority of them are benign in nature and can be dealt with in the postnatal period and most have a favourable outcome.^{1 2} Cysts arising from an undescended testis are extremely rare and pose a diagnostic and therapeutic dilemma.

This case highlights an intra-abdominal undescended testis presenting as an intra-abdominal cyst.

Torsion of the intra-abdominal testis is a relatively rare occurrence and the first case was reported by Gerster in 1898.¹⁰ Since then over 60 cases have been reported in literature (published in 2000) with malignancy being the most common predisposing condition for the torsion.^{10 11} The average age found with this condition is 20–40 years and the usual clinical presentation is abdominal pain with an empty hemiscrotum.^{10 12}

Perinatal testicular torsion (PTT) is a rare condition and is defined as torsion of the testis occurring within the first 30 days of life.¹

PTT presenting with a cystic abdominal mass is extremely rare and have only been reported a couple of times prior to this.^{6 7 9}

However, such a large cyst in a neonate has never been reported earlier. This diagnosis in the neonate poses a clinical dilemma as clinical examination and imaging may prove to be unequivocal.

The objective of presenting this case was to highlight the rarity of such an occurrence and also to bear in mind this condition as one of the differentials for an antenatally detected intra-abdominal cyst along with careful examination of the external genitalia.

Making the diagnosis of a torted, intra-abdominal undescended testis, especially in a neonate, is perplexing and requires a thorough physical examination to demonstrate the empty hemiscrotum and a tender abdominal mass. Histopathological examination alone can provide the final word on the diagnosis.

Patient's perspective

Translated from the patient's language:

- I am extremely privileged to be considered to contribute to the betterment of healthcare by permitting my son's condition to be published. I do understand my privacy will not be violated by this publication.

Learning points

- Undescended testes presenting as a cystic neonatal abdominal mass is extremely rare and torsion of this cystic mass presenting with an acute abdomen is rarer still.
- When considering the differential diagnosis of such a large cyst, testicular origin is hardly ever thought of and requires a thorough physical examination which includes the external genitalia to demonstrate the empty hemiscrotum.

Contributors I hereby confirm that I, FM, am the corresponding author. The second author is RBT from the Department of Pathology and I am grateful to him for the photomicrographs and interpretation of those to help substantiate the claims in this paper of an undescended testis. The third author is ISK, who is the head of the Unit of Paediatric Surgery, CMC, Vellore, and under whose guidance, the management of this case was done. All three authors have made substantial contributions to the conception and design of the work; the acquisition, analysis and interpretation of data for the work; drafting the work or revising it critically for important intellectual content and have given final approval of the version to be published. All are in

agreement to be 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. FM was responsible for the idea, conducting and reporting the case, planning the layout of the paper and analysis and revision of the paper. ISK was responsible for the idea, planning the layout of the paper, analysis, revision of the paper and for proofreading. RBT was responsible for proofreading and provided the histopathology photomicrographs and the interpretation of the photographs that were provided in the report. All three authors are guarantors of the paper. FM and IMK identified and managed the case.

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