Case Report

Two Cases of Fetal Inguinoscrotal Hernia and Review of the Literature

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Abstract

Fetal inguinal hernia is quite rare and here we report two cases of prenatally diagnosed inguinoscrotal hernia to add to the limited understanding of this rare condition. The disappearance of blood flow signal in the scrotum may be helpful in detecting fetal incarcerated inguinoscrotal hernia that may progress to strangulation. If bowel dilatation was observed in such cases, the physician should be alert to identify primary intestinal obstruction caused by congenital digestive tract malformation and secondary intestinal obstruction caused by incarceration.

Keywords: *Bowel dilatation, fetal inguinoscrotal hernia, incarcerated hernia, intestinal peristalsis*

Introduction

Although relatively common in pediatrics, an inguinal hernia is rarely diagnosed prenatally due to the incidence is probably lower than after birth and the physical examination of the fetus is not possible. Sixteen antenatal diagnosed cases have been reported in English literature from 1991 to 2016. We report two cases of prenatally diagnosed fetal inguinoscrotal hernia from Women's Hospital, Zhejiang University School of Medicine, diagnosis was confirmed and laparoscopic herniorrhaphy of hernia was performed without complications postnatally. This article focuses on prenatal imaging features, differential diagnosis, and pregnancy management, the significance of abnormal bowel peristalsis, the disappearance of blood flow signal, and bowel dilatation in fetal inguinoscrotal hernia are discussed as well.

Case Presentation

Case 1

A 32-year-old primigravid woman, gravida 2 para 0, with a history of previous first-trimester abortion, who underwent *in vitro* fertilization resulting in a dichorionic/diamniotic twin gestation, was referred to our unit at 21 weeks gestation due to suspected fetal anomaly in Twin 2. Multiple malformations was identified in Twin 2 (female) while Twin 1 (male) had normal anatomy. The patient opted for selective reduction of Twin 2 and feticide was achieved using a direct intracardiac injection of potassium chloride (1.5 g in 10 mL). Amniocentesis performed simultaneously in Twin 1 revealed a normal 46 XY karyotype. At 39+3 weeks' gestation, sonographic examination revealed a 4.6*3.5*3. cm right scrotal mass with a homogeneous echogenicity and regular wall. With the diagnosis of a scrotal hemorrhage versus tumor, a further ultrasound examination was performed and the mass turned to be nonhomogeneous in echotexture and predominantly solid with few small echo-free cystic areas, in which Power Doppler showed a few signals of blood flow [Figure 1a]. Echo movements within the mass suggestive of bowel peristalsis were seen in the scrotal mass. Dilated bowel loop measuring 16 mm in width was observed without thickened bowel wall. An MRI assessment was undertaken on the same day. On MR image, the material had a signal intensity similar to that of small bowel that extended from the abdominal cavity into the scrotum on both fast imaging employing steady-state acquisition (FIESTA) and liver acquisition

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with volume acceleration-flexible (LAVA-Flex) T1-weighted [Figure 1b]. The right scrotum was significantly larger than the left [Figure 1c]. There was no imaging evidence of associated bowel obstruction, meconium peritonitis, ascites or volvulus, abdominal calcifications, or an intra-abdominal mass lesion. Fetal biophysical parameters were compatible with the age of gestation and the amount of amniotic fluid was considered to be normal. Based on sonographic and MRI visualization, the diagnosis of a fetal inguinoscrotal hernia without incarceration or strangulation was made.

A pediatric surgeon counseling was requested which recommended clinical evaluation immediately after birth and emergency surgery in case of complications. An ultrasonography was taken on the next day and bidirectional bowel peristalsis within the scrotum was detected which may be suggestive of intestinal obstruction and incarcerated hernia. The patient had an

Table 1: Fetal inguinoscrotal hernia. Review of the literature

First author	GA	Side	Size (cm)	Other anomalies	Bowel peristalsis	Blood flow signal	Bowel dialation	Neonatal outcome
Kithir,[4] 2016	37	Right	2.9*1.7	Not described	Present	NA	Not described	spontaneous regression after birth
Ronzoni,[1] 2015	31	Right	NA	complete fetal AV block, low anorectal malformation with a perineal fistula	Present	NA	Present 13–17 mm in width	Operated after 6 months
Massaro,[5] 2013	37	Right	5.4*4.6	Not described	Present, delayed	Present	Not described	Operated 10 days
Khatib,[6] 2013	36	Bilateral	4.2*3/3.2*2.8	Not described	Present	Present	Not described	Operated 1 month
	24	NA	NA	Not described	Present	NA	Not described	Operated 1 month
Basaran,[7] 2010	28	Left	NA	Multiple anomalies (spondylocostal dysostosis)	Present	NA	Not described	Died 3 days
Frati,[8] 2008	35	Right	5.5	Not described	Present	Present	Not described	Operated 4 days
Caserta,[9] 2007	21	Right	3.3*3	Not described	Present, delayed	Absent	Not described	Operated after birth
Ji,[10] 2005	36	Left	4.2*5*3.8	multiple joint contractures	Absent	Present	Not described	Died 5 days
Allen,[2] 2004	34	Left	3.6*3	Cystic fibrosis	Present	NA	Present	Operated 3 weeks and right IH repaired
Sharma,[3] 2004	34	Right	3.2*2.7	Not described	Absent	Absent	Not described	Operated 3 days
Kesby,[11] 1997	36	Right	4.6*5.1*4.3	Not described	Present	Absent	Not described	Operated 15 and 28 days due to left IH
Paladini,[12] 1996	30	Right	4.2	Trisomy 18, multiple anomalies (omphalocele, clubfeet, clenched hands)	Present, delayed	NA	Not described	Died 3 hours after birth
Shipp,[13] 1995	37	Right	4	left-sided hydrocele with torsion of the left testis.	Present	Present	Not described	Operated in neonatal period
Meizner,[14] 1992	33	Right	6.5*5.6*5.4	Not described	Present	NA	Not described	Reduction after birth and operated 7 days

GA=gestational age, NA=not available, MRI=magnetic resonance imaging; IH

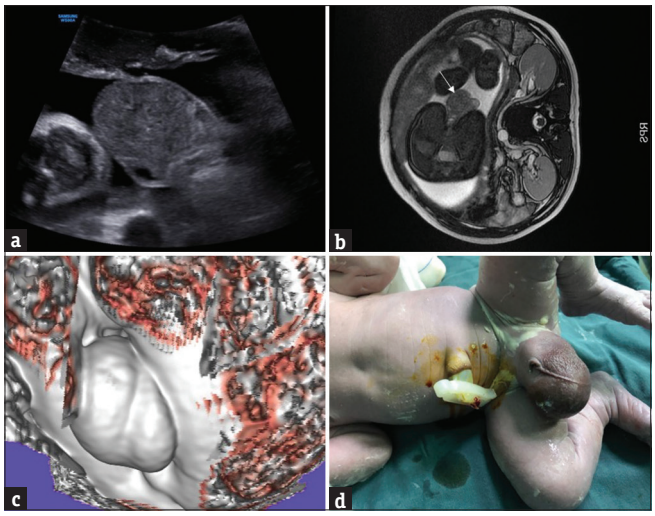

Figure 1: (a) Transverse ultrasonographic image of the fetal scrotum; (b) Bowel loop in the right scrotum displayed hypointense signals on T2WI (white arrow); (c) The 3-dimensional volume render (3DVR) image vividly showed the right scrotum was significantly larger than the left; (d) The appearance at birth

emergency cesarean section delivery for the prevention of strangulation secondary to incarceration. A male infant weighing 2720 g was delivered with APGAR scores of 10 and 10 at one and five min, respectively. The infant was noted to have a right-sided inguinoscrotal hernia, approximately 5 * 3 cm in size without overlying skin changes [Figure 1d]. Postnatal doppler assessment of the mass showed an absence of blood flow in the scrotum and bidirectional bowel peristalsis become more obvious. A hernia manipulative reduction of the incarcerated inguinoscrotal hernia was performed three hours after birth. The inguinoscrotal hernia recurred at six months old during the follow-up period because the parents refuse surgical correction of the hernia. Finally, an uncomplicated laparoscopic herniorrhaphy was performed at age of eight months. At the time of this report, the baby is thriving and there were no bowel symptoms on postoperative follow-up at age of 10 months.

Case 2

A 28-year-old woman, gravida 1 para 0, was referred at 36 weeks of gestation for an anatomical survey of the fetus and a high suspicion of a testicular tumor. The obstetrical history was unremarkable. We could observe an enlarged scrotum with an echogenic mass inside, bowel peristalsis within the scrotum, and a few signals of blood flow were detected [Figure 2a]. On the MR image, the axial liver acquisition with volume acceleration (LAVA) sequence showed that the right scrotum was filled with small bowel which presented as hyperintense [Figure 2b]. Based on the sonographic visualization, the diagnosis of an inguinal-scrotal hernia

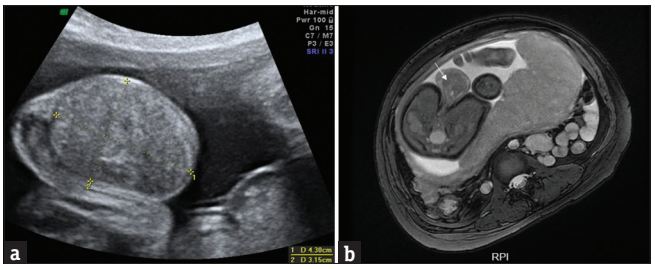

Figure 2: (a) Transverse ultrasonographic image of the fetal scrotum; (b) Bowel loop in the right scrotum displayed hypointense signals on liver acquisition with volume acceleration-flexible (LAVA-Flex) T1-weighted

was made. At 38+3 weeks gestation, the patient delivered vaginally a 3200 g male infant with Apgar scores of 10 and 10 at one and five minutes, respectively. The inguinoscrotal hernia was confirmed after birth and the surgical correction was performed at age of one month uneventfully.

Discussion

In this paper, we report two cases of prenatal diagnosed incarcerated inguinal scrotal hernia and one case occurs in the remaining co-twin after selective fetal reduction in dichorionic twins. Case 1 represents a combination of fetal findings including abnormal bowel peristalsis, bowel dilatation, and incarcerated inguinoscrotal hernia. This constellation of findings raises two questions,: (1) What is the significance of abnormal bowel peristalsis for fetal inguinoscrotal hernia? (2) Is bowel dilatation related in some way to fetal inguinoscrotal hernia—can it be primary or secondary bowel obstruction?

Congenital inguinal hernia is a relatively common finding among neonates and infants with an incidence of 0.88% to 4.4% at birth and 9% to 11% of those born prematurely.[1] There is male preponderance in the prevalence of inguinal hernia.[2,3] Conversely, the prenatal inguinoscrotal hernia is a rare condition and we only found 16 antenatal diagnosed cases in English literature from 1991 to 2016 [Table 1], in which 15 cases were singleton pregnancies and one case was a triplet gestation after selective reduction.

The right side is affected more commonly with approximately 62.5% (10/16) on the basis of reported data. Although pediatric inguinal hernia is usually an isolated finding, several associated malformations were reported in prenatally diagnosed cases including trisomy 18 with omphalocele,[12] imperforate anus with recto-vesicle fistula,[1] cystic fibrosis,[2] or syndromes (Jarcho-Levin syndrome or unidentified syndrome).[7,10]

Fetal inguinoscrotal hernia typically appears in the third trimester as a complex scrotal mass detected by obstetric

sonography. Echogenic structures consistent with bowel loops within the scrotum, predominantly solid echogenic content with internal echo-free cystic components, support the diagnosis. If ultrasound can't determine the nature of scrotal contents, MRI could be conducted for a definitive diagnosis and show the signal intensity similar to that of small bowel on both longitudinal relaxation time– and transverse relaxation time-weighted imaging, continuing from the abdominal cavity into the scrotal sac.[10] The most sensitive sonographic characteristic of fetal inguinoscrotal hernia are visualization of bowel peristaltic waves within the scrotum, which is considered the pathognomonic feature.[11] In our case, peristaltic waves were identified at subsequent ultrasound examination while we found a smooth contour mass with nonhomogeneous components and no bowel peristalses during the first scan. The appearance of peristaltic waves was also delayed in a fetus with trisomy 18 and omphalocele, which can be interpreted by the fact that most of the bowel loop herniated into the omphalocele.[12] Physicians should perform sonographic re-assessment at a later stage after failling to demonstrate bowel peristalsis in strongly suspected cases of inguinoscrotal hernia, owing to delayed bowel peristalsis not seen at the time of examination and may appear at a later stage sometimes. Sharma *et al*. reported a case without bowel peristalsis within the scrotum but confirmed as a nonreducible inguinoscrotal hernia with edematous small bowel in it.[3] The author suggested that the eliminated bowel peristalsis was caused by the edematous small bowel contained within the herniated sac which allows slow passage of bowel content with minimal peristalsis. However, the absence of bowel peristalsis does not rule out a fetal inguinoscrotal hernia in cases of inguinal scrotal hernia with strangulation, bowel obstruction, or ischemia.[11]

The key factor in the development of inguinoscrotal hernia is thought to be the increasing intra-abdominal pressure such as vigorous crying, prematurity, chronic lung disease, ascites, and bowel pathology after birth, which force bowel loops to pass through the abdominal wall into the scrotum.[11] However, the factors leading to increased intra-abdominal pressure during fetal life remain unknown, which is similar to the intra-amniotic pressure. In theory, The sudden abdominal pressure increasing in utero may lead to an incarcerated hernia, and long-term incarceration may lead to a blood supply disorder and strangulated hernia. In our case, after the first observation of normal intestinal peristalsis, a bidirectional intestinal peristalsis wave was observed the next day and there was no sonographic evidence to indicate an associated ascites, abdominal mass, and thickening intestinal wall. The question was raised of whether the unusual pattern of bowel peristalsis was secondary to the hernia incarceration. Since it was also observed after birth, we suggest a bidirectional intestinal peristalsis wave could be a sign for the early stage of incarceration in a prenatal inguinoscrotal hernia. It is not important of the pathological significance of blood flow within the mass. Usually, there is a paucity of blood flow on color Doppler examination, but occasionally blood flow signals originating from the mesenteric artery may be seen within the mass which demonstrates that the mass is a non-strangulated inguinal scrotal hernia.[8,10]

Primary or secondary intestinal obstruction may lead to prenatal intestinal dilatation. Two published cases reported an association between bowel dilatation or bowel obstruction with a fetal inguinoscrotal hernia so far. Intestinal dilatation was presented in one case with anorectal malformation and another case with cystic fibrosis.[1,2] The authors hypothesized that hernia is secondary to increased intra-abdominal pressure caused by primary bowel obstruction during fetal life. We also showed bowel loop dilatation in our case. Therefore, if bowel dilatation was observed in the case of a fetal inguinoscrotal hernia, it is necessary for the physician to check whether there is congenital digestive tract malformation and be alert to the secondary intestinal obstruction caused by incarcerated hernia.

The significance of the prenatal diagnosis of fetal inguinal hernia lies in the obstetric management, clinical evaluation at birth, and follow-up in neonates. The mode and timing of delivery should be discussed based on obstetrical indications. Spontaneous regression of uncomplicated fetal inguinoscrotal hernia during the perinatal period is a possible outcome.[4] A timely diagnosis allows concerns to be discussed between the parents and care providers before delivery, which may alleviate stress and confusion in the immediate postnatal period. Although there is no report of prenatal strangulated hernia, our case suggested that incarcerated inguinoscrotal hernia is possible. After the diagnosis of fetal inguinoscrotal hernia is made, serial ultrasonographic surveillance should be provided to follow both the signs of obstruction such as dilated bowel loops and polyhydramnios, and the change of bowel peristalsis. If features of obstruction and incarceration are present and the fetus is mature, timely delivery should be planned to avoid intestinal necrosis caused by long-term incarceration.[11] In cases of uncomplicated fetal inguinoscrotal hernia, usually, surgical repair will be carried out shortly after diagnosis, as there is a 30% to 40% risk of incarceration and possible strangulation in the first year of life.[4] Early detection of inguinal hernia incarceration allows a successful manual reduction in our

case. As the complication rate of elective herniorrhaphy was lower than that of emergency operation, an elective laparoscopic operation was performed at age of three months without complications.

In conclusion, the Identification of fetal inguinal hernia allows for obstetric management, clinical evaluation at birth, and follow-up in neonates. when a scrotal mass is identified in utero, it is important to evaluate all ultrasonographic signs of the lesion to discriminate inguinoscrotal hernia from other scrotal masses. Clinicians should be careful in patients with abnormal bowel peristalses and sonographic evidence indicating secondary intestinal obstruction such as bowel dilatation, abdominal mass, associated ascites, and thickening intestinal wall. It is thought that bidirectional intestinal peristalsis could be a sign of bowel incarceration in prenatal inguinoscrotal hernia, and termination of pregnancy can effectively avoid strangulation caused by long-term incarceration. Due to the complication of emergency operation and the recurrence risk, elective herniorrhaphy was recommended after birth.

Declaration of patient consent

The authors certify that they have obtained the appropriate patient consent form.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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