

CASE REPORT

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Antenatal diagnosis and management of fetal inguinoscrotal hernia: A case report with literature review

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Abstract

Primary inguinal hernia is a common pediatric surgical condition with an incidence of 1%–4%, which is higher in male or premature newborns. It is characterized by the protrusion of abdominal contents through inguinal canal in newborns. However, prenatal fetal inguinal hernia is a rare condition because the pressure of amniotic fluid is similar to intra-abdominal pressure. Only 19 English publications were found with 21 reported cases until now. We report belatedly discovered inguinoscrotal hernia at 38⁺⁰ weeks' gestation. Usually, the fetal testicular descent begins from 24 to 25 weeks' gestation, and it is found after 32 weeks of gestational age in 97% of the fetuses. Therefore, it is necessary to get into the habit of checking fetal testicles during routine US after at least 32 weeks of gestational age.

KEYWORDS

fetal inguinal hernia, fetal inguinoscrotal hernia, scrotal mass

1 | INTRODUCTION

As advanced prenatal ultrasonography becomes widely used, most major fetal anomalies can now be detected. While major anomalies, such as brain or heart anomalies are relatively easy to suspect and detect, fetal inguinal hernias are challenging to predict and can easily be overlooked.

A systematic search of Pubmed, Science Direct, and Springer databases was conducted to identify articles and case reports based on the keywords and phrases “fetal inguinal hernia,” and “inguinoscrotal hernia.” The search was limited to English articles. After Ober and Smith first described fetal inguinoscrotal hernia in 1991, only 19 publications were found, reporting 21 cases up to now.¹

Primary inguinal hernia is a common pediatric surgical condition with an incidence of 1%–4%, which is higher in male or premature newborns.^{2,3} It is characterized by the protrusion of abdominal contents through inguinal canal in newborns. However, prenatal fetal inguinal hernia is a rare condition because the pressure of amniotic fluid is similar to intra-abdominal pressure during pregnancy.⁴

Therefore, it is easy to miss, as most physicians forget to follow up the genital area after mid-trimester screening ultrasonography.

We present a case report that was almost missed but was belatedly discovered at 38⁺⁰ weeks' gestation, diagnosed as an inguinoscrotal hernia. In addition to conducting the mentioned databases-based analysis of the antenatal diagnosis and management of fetal inguinoscrotal hernia, we intend to analyze and provide an overview of the reported cases.

2 | CASE REPORT

A 38-year-old primigravida, gravida 1 para 0, who conceived after third cycle of in vitro fertilization, first visited to Chung-Ang University Gwangmyeong Hospital at 16⁺⁰ weeks' gestation for a routine fetal check-up. She was a carrier of HBV and had a history of prior surgeries, including pelviscopic left salpingectomy due to ectopic pregnancy and hysterocopic endometrial polypectomy. Before her visit to our hospital, routine obstetric ultrasound examinations and

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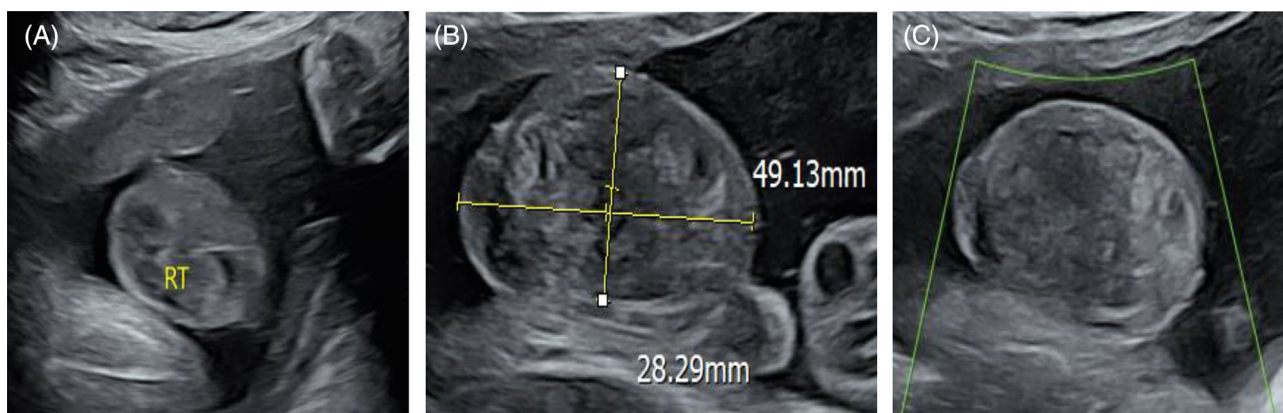
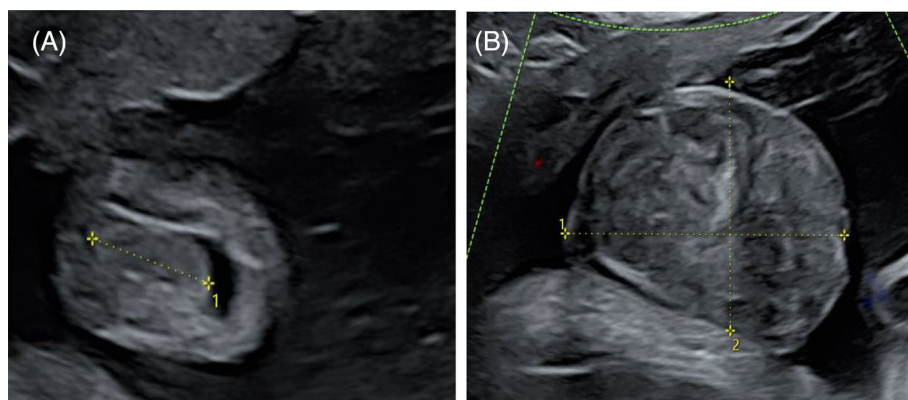


FIGURE 1 The findings of inguinoscrotal hernia initially detected at the 38-week ultrasound. (A) Right scrotum appears normal. (B) Left scrotum suspected of a scrotal mass measuring 4.9×2.8 cm. (C) No Doppler flow on left scrotum.

FIGURE 2 Fetal scrotum at 39-week. (A) Right scrotum with a small amount of hydrocele. (B) No change in the size of the left scrotal mass. We observed peristalsis during the examination.



laboratory findings in the first and second trimesters revealed no abnormalities. At 20⁺¹ weeks' gestation, a mid-trimester screening ultrasound was performed, confirming a normal male fetus.

At 38⁺⁰ weeks' gestation, we incidentally discovered an enlarged left scrotum in the fetus, measuring 4.9×2.8 cm, during a routine ultrasound examination (Figure 1). Upon initial discovery, we suspected a tumor mass in the left scrotum due to its complex echogenicity and absence of color Doppler-positive vessels. Peristalsis was not clearly recognized during this examination. The right scrotum and testis appeared normal in size. One week later, a follow-up ultrasound examination was performed before induction of labor. The size of the intra-scrotal mass remained unchanged, and peristalsis was observed during the examination (Figure 2, Video clip 1). As a result, we became confident that the abnormal echogenic mass was indicative of fetal inguinoscrotal hernia.

At 39⁺¹ weeks' gestation, an emergency Cesarean section was performed due to failure to progress. Postoperatively, the mother underwent embolization because of massive bleeding associated with placenta accrete and uterus atony. The delivered infant, a 3000 g male, had Apgar score of 9 and 9 at 1 and 5 min, respectively. On the second day after birth, the infant genitalia ultrasound revealed a left inguinoscrotal hernia containing intact small bowel and intact right scrotum. A pediatric surgeon performed inguinal herniorrhaphy, a

surgical correction, on the fourth day after birth without any complications. After 1 week after surgery, the infant was discharged uneventfully. However, the infant presented with continuous projectile vomiting and poor weight gain, leading to a diagnosis of hypertrophic pyloric stenosis. On the 28th day after birth, pyloromyotomy was performed.

3 | DISCUSSION

Inguinal hernia, specifically an indirect hernia, is a common congenital condition associated with an intestinal hernia occurring through the widely patent processus vaginalis within the inguinal canal. While theoretically possible in both males and females, congenital inguinal hernia is observed to be 10 times more common in males.^{5,6} The overall incidence of inguinal hernia in childhood ranges approximately from 0.8% to 4.4%, with a higher prevalence of 9%–11% in premature infants compared to term infants.⁷ In male, the diagnosis of inguinoscrotal hernia is made when intestines enter the canal anterior to the spermatic cord, extending through the external inguinal ring into the scrotum. In infants, inguinal hernia may occur if the processus vaginalis persists, and increased intra-abdominal pressure during crying leads to the protrusion of bowel loops into the scrotum.⁸

TABLE 1 Fetal inguinoscrotal hernia.

Primary Author (year) [Reference number]	Maternal age	GA at diagnosis	Hernia side	Size (cm)	Blood flow signal	Bowel peristalsis	Other abnormal prenatal findings	Prenatal MRI	Method of delivery	GA at delivery	Weight at delivery (g)	Outcome of the hernia
Liberty et al. (2023) ¹⁹	34	29	Bilateral	4.4 × 2.5 × 2.7	Mild	(+)	Hydrocele	NA	C/sec	39	3900	Surgical correction after 3 weeks
Chen et al. (2022) ⁷	32	39 + 3	Right	4.6 × 3.5 × 3	Few	(+)	R/O intestinal obstruction and incarcerated hernia	(+)	C/sec	39 + 4	2720	Surgical correction after 8 months
	28	36	Right	NA	Few	(+)	NA	(+)	VD	38 + 3	3200	Surgical correction after 1 month
VS Kumar et al. (2020) ¹⁸	19	35	Right	3.8 × 3.6 × 2.5	(+)	(+)	Fetal growth restriction (<1 percentile) Confirmed Trisomy 18	NA	VD	36	1500	Fetal demise at GA 36 weeks
Kithir et al. (2016) ¹⁷	22	37	Right	2.9 × 1.7	NA	(+)	Hydrocele	NA	VD	39	3050	Spontaneous regression
Ronzoni et al. (2015) ¹⁶	31	32	Right	NA	NA	(+)	Dilated bowel loop, low anorectal malformation with a perineal fistula	NA	C/sec	35	2230	Anoplasty after 2 days Surgical correction after 6 months
Khatib et al. (2013) ¹⁵	29	36	Bilateral	4.2 × 3 3.2 × 2.8	(+)	(+)	NA	NA	VD	39	3160	Surgical correction after 1 month
	25	24	NA	NA	NA	(+)	NA	NA	NA	NA	NA	Surgical correction after 1 month
Massaro et al. (2013) ⁴	30	37	Right	5.4 × 4.6	(+)	(+) delayed	Hydrocele	NA	C/sec	39	3700	Surgical correction after 10 days
Basaran et al. (2010) ¹⁴	28	33	Left	NA	NA	(+)	Spondylocostal dysostosis	NA	VD	40	2900	Died in 3 days
Fрати et al. (2008) ¹³	29	35	Right	5.5	(+)	(+)	NA	NA	VD	40	3650	Surgical correction after 4 days
Caserta et al. (2007) ¹²	29	21	Right	3.3 × 3	(-)	(+) delayed	NA	NA	C/sec	37	3300	Surgical correction after 7 days
Ji et al. (2005) ⁶	30	36	Left	4.2 × 5 × 3.8	(+)	(-)	Multiple joint contractures (both knees, right first carpometacarpal, left foot)	(+)	C/sec	37	3200	Died in 5 days
Allen et al. (2004) ¹¹	26	34	Left	3.6 × 3	NA	(+)	Cystic fibrosis	NA	VD	NA	2676	Surgical correction after 3 weeks
Sharma et al. (2004) ⁵	30	34	Right	3.2 × 2.7	(-)	(-)	NA	NA	C/sec	36	2460	Surgical correction after 3 days

TABLE 1 (Continued)

Primary Author (year) [Reference number]	Maternal age	GA at diagnosis	Hernia side	Size (cm)	Blood flow signal	Bowel peristalsis	Other abnormal prenatal findings	Prenatal MRI	Method of delivery	GA at delivery	Weight at delivery (g)	Outcome of the hernia
Kesby et al. (1997) ¹⁰	28	36	Right	4.6 × 5.1 × 4.3	(-)	(+)	NA	NA	VD	39-40 ^a	3580	Surgical correction after 13 days
Paladini et al. (1996) ⁸	26	30	NA	4.2	NA	(+)	Confirmed Trisomy 18, Multiple anomalies (AVSD, omphalocele, clubfeet, clenched hands)	NA	VD	37	2300	Died in 3 h after birth
Shipp and Benacerraf, (1995) ³	30	40	Right	4	(+)	(+)	Left side hydrocele, torsion of the left testis	NA	VD	40	NA	Surgical correction and excision in the neonatal period
Hussain and Doiron (1995) ⁹	32	41	Bilateral	11 × 9 × 8	(-)	(+)	NA	NA	C/sec	41	4096	Surgical correction after 2 days
Meizner et al. (1992) ²	21	33	Left	6.5 × 5.6 × 6.4	NA	(+)	NA	NA	VD	40	3400	Surgical correction after 7 days
Ober and Smith (1991) ¹	23	41	Right	5.9 × 4.8	NA	(+)	(-)	NA	VD	41-42 ^b	NA	Surgical correction after 1 day

Note: Review of the literature.
 Abbreviations: AVSD, atrioventricular septal defect; C/sec, Cesarean section; GA, gestational age; NA, not applicable; R/O, rule out; VD, vaginal delivery.
^aThe following day of 39 weeks' gestation.
^b2 days later of 41 weeks' gestation.

However, fetal inguinoscrotal hernia is a rare condition during the antenatal period due to the similar pressure of amniotic fluid and fetus intra-abdominal pressure in the uterus. Consequently, only 19 English publications with 21 reported cases have been found since 1991 through a systemic search of NCBI PubMed, Science Direct, and Springer and these have been summarized in Table 1.^{9–19}

Fetus testicular descent is a complex process and divided into two stages; the transabdominal stage and the inguinoscrotal stage. The transabdominal stage occurs with non-androgenic hormone (Leydig insulin-like hormone) between the eighth and fifteenth gestational weeks, progressing from the posterior abdominal wall to the internal inguinal ring. Traction is exerted by the gubernacular bulb anchored in the scrotum. The inguinoscrotal stage, conversely, commences at the 26th gestational week, progressing from the internal inguinal ring to the scrotum and is primarily controlled by androgen.²⁰ However, only 7% and 12.5% of the testes descend at 25th gestational weeks and 27th gestational weeks, respectively according to the Table 1. More than 95% of the fetal testes descend into the scrotum by the 33rd gestational week. Consequently, it is necessary to check whether the testes have descended at least once after 33rd week of pregnancy. Moreover, the majority of inguinoscrotal hernia occur in the late third-trimester of pregnancy, based on reported cases until now, with only two exceptions.^{12,14}

The most common sonographic feature suggesting the diagnosis of fetal inguinoscrotal hernia is the presence of multiple echocystic masses within fetal scrotum. Real time ultrasound allows the observation of wriggling movements, indicating bowel peristalsis. Normal scrotal contents may or may not be visible on ultrasound. Inguinoscrotal hernia is a form of indirect hernia, and it is noted that indirect hernias more frequently occur on the right side. In this study, 12 cases occurred on the right side, believed to be attributed to the slower closure of the patent processus vaginalis on the right side compared to the left. According to the Table 1, three cases underwent prenatal MRI to confirm the diagnosis.^{6,7} However, it is not routinely recommended to perform prenatal MRI for a more accurate diagnosis.

Even if no anomaly findings are observed on mid-trimester screening ultrasonography, it is imperative to reevaluate for other structural anomalies when inguinoscrotal hernia is discovered. According to the Table 1, the most common prenatal ultrasound finding associated with inguinoscrotal hernia is hydrocele. Among the 21 fetuses, three exhibited structural anomalies, including low anorectal malformation with a perineal fistula, spondylocostal dysostosis (SCDO) and multiple joint contractures. Notably, SCDO is already known to be related to inguinal hernia.²¹ Furthermore, two fetuses were diagnosed with Trisomy 18 during prenatal period, presenting with severe fetal growth restriction (estimated fetal weight < 1st centile) or multiple anomalies such as atrioventricular septal defect, omphalocele, clubfeet, and clenched hands. In summary, inguinoscrotal hernia may be associated with other structural anomalies or chromosomal abnormalities. Therefore, if inguinoscrotal hernia is suspected, a targeted ultrasound examination should be conducted slowly and meticulously to ensure there are no other anomalies.

However, a routine invasive test, is not deemed necessary unless other structural abnormalities coexist with inguinoscrotal hernia.

Anomalies that need to be differentiated from inguinoscrotal hernia include the presence of tumors that can occur in the perineal region, such as sacrococcygeal teratoma and lipoma.^{10,19} These tumors can be distinguished through ultrasound findings, considering the presence or absence of color Doppler flow or bowel peristalsis. Evaluating the characteristics and blood flow of the tumor is crucial for making an accurate diagnosis.

Moreover, if fetal inguinoscrotal hernia is suspected, it is essential to conduct a thorough examination for potential complications such as strangulation or bowel obstruction. However, the incidence of such complications is very low, and as of now, there have been no occurrences in cases of inguinoscrotal hernia.

The timing of delivery is not strongly associated with the presence of inguinoscrotal hernia, and as summarized in Table 1, the majority of cases delivered after 37 weeks. Excluding infants with Trisomy 18 and multiple anomalies and as well as one who experienced spontaneous regression after birth, all other infants received successful surgical intervention and had no specific issues. The median interval to surgery was 13 days after birth.

4 | CONCLUSION

Fetal inguinoscrotal hernia, although uncommon, should not be overlooked in prenatal examination, as its detection and diagnosis are not challenging. Especially in the case of male fetuses, it is recommended to develop a habit of re-examining the genitalia at least once after 33 weeks of gestation. Furthermore, potential complications such as strangulation, bowel obstruction, and torsion should be considered despite their rarity. Depending on the suspected occurrence of these complications, a delivery plan should be established.

AUTHOR CONTRIBUTIONS

Conceptualization: Seong JS, Kim YM. *Data curation:* Seong JS, Nam GN. *Formal analysis:* Seong JS, Kim M. *Writing-original draft:* Seong JS. *Writing-review & editing:* Seong JS, Kim YM, Nam GN, Kim GJ.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that supports the findings of this study are available in the supplementary material of this article.

ETHICS STATEMENT

Informed consent was obtained from the patients and the requirement of IRB was waived.

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SUPPORTING INFORMATION

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