



Incarcerated uterus and bilateral ovaries in a premature female infant inguinal hernia

Elizabeth A. Marshall, Scarlett B. Hao, David A. Rodeberg^{*}

Viridant Medical Center, East Carolina University Department of Surgery, Division of Pediatric Surgery, USA

ARTICLE INFO

Keywords:

Inguinal hernia
Uteri inguinale
Fallopian tube
Female
Prematurity

ABSTRACT

We present a case of an inguinal hernia in a premature female infant containing the uterus and bilateral adnexa. This was diagnosed on ultrasound and underwent operative repair. The viable hernia contents were reduced laparoscopically, however the sliding nature of the hernia warranted open repair. Surgery was converted to the traditional inguinal approach and the hernia was successfully repaired with high ligation of the sac and additional closure of the internal ring. Post-operative course was unremarkable.

The most common congenital anomaly in pediatric patients is the indirect inguinal hernia, affecting males more than females, and affecting premature infants more than infants born full term [1,2]. In female patients, the most common herniated organ is the ovary, seen in up to 20% of patients, occasionally involving the fallopian tube [3]. It is especially rare, however, to find both the uterus and adnexa in a unilateral inguinal hernia. We report on a six-week-old female infant born 31 weeks premature with an inguinal hernia containing uterus and bilateral ovaries.

1. Case report

A six week old infant girl who was 37 weeks gestational age presented to a local hospital with a new bulge in the right groin. An ultrasound was performed (Fig. 1) and visualized both ovaries and uterus within the inguinal hernia. The infant was immediately transferred to our tertiary care facility. On initial exam, the hernia was not reducible, however the patient was not in any distress. A repeat Doppler ultrasound (Fig. 2) was acquired which demonstrated good blood flow to all structures, so repair was scheduled for first case in the morning—a waiting period of approximately 6 h. The patient subsequently underwent laparoscopic reduction of the viable hernia contents (Fig. 3). The hernia had a portion of the fallopian tube comprising the hernia wall (Fig. 4), therefore given the sliding component of the hernia, the decision was made to convert to an open procedure. A traditional right inguinal incision was made, allowing isolation of the hernia sac with subsequent dissection and reduction of the fallopian tube. We performed

a high ligation of the hernia sac with additional suture closure of the internal ring. The patient had an uneventful postoperative course with adequate pain control, tolerance of oral feeds, and was discharged home on day of operation. Patient had no complications present at the three-week follow-up visit.

2. Discussion

There are a few case reports describing diagnosis and repair of indirect inguinal hernias containing uterus, fallopian tubes and both ovaries in infants [4–13]. The majority of these reports advocate ultrasound as the diagnostic test of choice, though the technique and timing of the repair has varied [4–6]. While ultrasound has been shown to consistently identify the presence of the uterus in the inguinal hernia, it can fail to identify involvement of the ovary [7–9]. This discovery is often noted at the time of surgical exploration. Doppler ultrasound, in conjunction with clinical exam, is a reliable method to identify strangulation, a factor that determines if surgical correction is potentially emergent [5]. Some surgeons have delayed repair up to nine weeks, whereas others have operated on an emergent basis. Delay of surgical repair did not affect viability of hernia contents in the absence of signs of strangulation, despite evidence that inguinal hernias containing an ovary are at increased risk of incarceration compared to those that contain bowel only [6]. Of these cases, several surgeons chose to explore the contralateral inguinal region at time of open surgical intervention, often finding a hernia defect that was then repaired during the same operation [4,7,8,13]. This finding is supported by a population study

^{*} Corresponding author. 600 Moye Boulevard, PCMH - MA, Room 207, Greenville, NC, 27834, USA.

E-mail address: rodebergd@ecu.edu (D.A. Rodeberg).

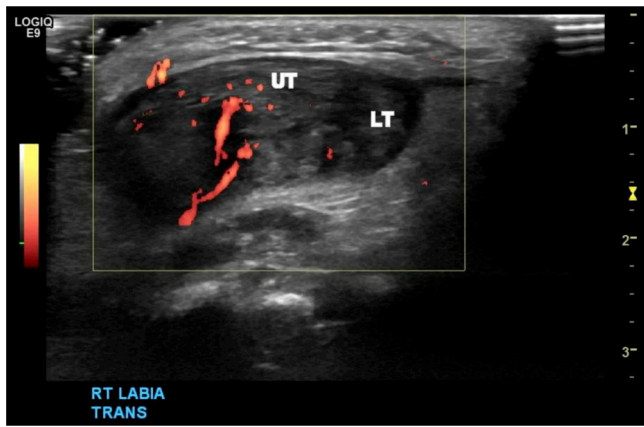


Fig. 1. Ultrasound of hernia contents with Doppler flow showing perfusion. Abbreviations: UT, uterus; LT OV, left ovary.

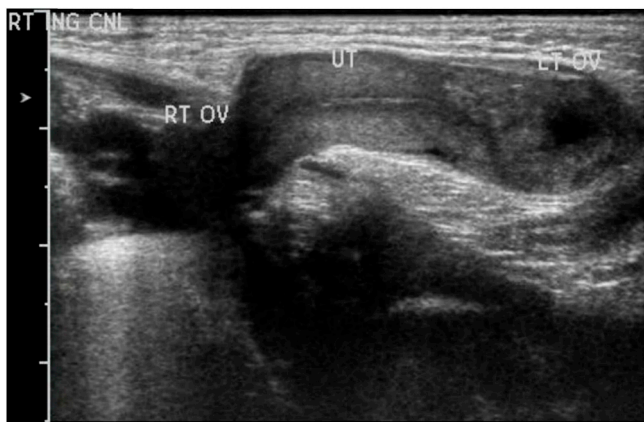


Fig. 2. Transverse view of ultrasound of hernia contents. Abbreviations: RT OV, right ovary; UT, uterus; LT OV, left ovary.

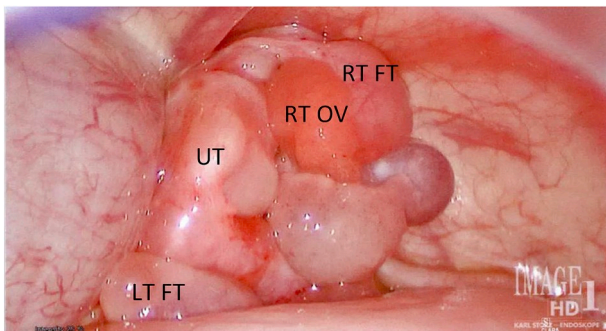


Fig. 3. Laparoscopic view of bilateral ovaries, tubes, and uterus at the right inguinal internal ring. Abbreviations: RT FT, right fallopian tube; RT OV, right ovary; UT, uterus; LT FT, left fallopian tube.

published in 2016 by Chang et al., which concluded that contralateral concurrent inguinal hernias occurred in 25% of females [14]. Another study by Burgmeier et al. revealed that the rate of contralateral patent processus vaginalis in premature female infants was as high as 76.9% [15].

Because of the likelihood of finding a contralateral defect, some have advocated contralateral exploration as part of a standard approach [8]. An initial laparoscopic approach has the combined benefit of reduction and evaluation of the hernia content viability as well as evaluation of the contralateral side. This approach is especially suited to cases of

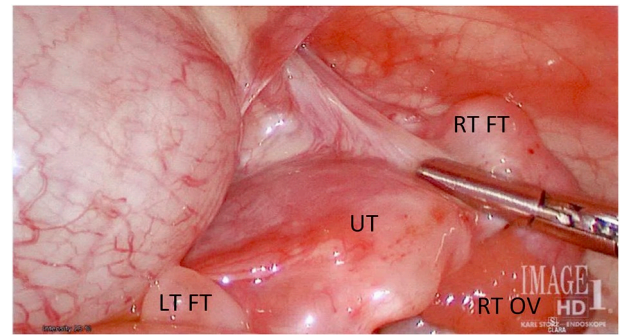


Fig. 4. Laparoscopic view of hernia contents reduced into the abdominal cavity with sliding fallopian tube. Abbreviations: RT FT, right fallopian tube; RT OV, right ovary; UT, uterus; LT FT, left fallopian tube.

incarcerated hernias [16]. Since the introduction of a laparoscopic approach to pediatric inguinal hernia repair two decades ago with intracorporeal sac ligation, techniques have developed to also enable laparoscopic internal ring closure using an extracorporeal approach [17]. In the eighteen published cases of inguinal hernia containing uterus and bilateral adnexa, open repair with ring closure was performed in 73% of patients [4–13]. None of these cases reported recurrence of hernia, with follow-up time ranging from eight weeks to sixteen months.

It is known that indirect inguinal hernia defects in female infants occur due to a persistence of the Canal of Nuck, which usually closes cephalocaudally between eight months gestation and birth [5,6]. This explains the increased risk of inguinal hernia present in premature infants [1,2,10]. Indirect inguinal hernias containing uterus, fallopian tubes and bilateral ovaries were twice as likely to occur on the left side compared to the right [4–10,12,13]. This is congruent with the findings of a population study by Burgmeier et al. that discovered left sided inguinal hernias occurred at more than double the rate of right sided inguinal hernias in premature female infants [15]. No anatomical explanation has been found for this phenomenon. However, the predominance of left sided inguinal hernias containing uterus and bilateral adnexa and indirect inguinal hernias in premature infants may suggest an incompletely synchronous bilateral obliteration of the Canal of Nuck.

The existing literature suggests that the uterus, fallopian tubes, and bilateral ovaries end up in the hernia sac due to a sliding hernia mechanism, whereby the peritoneum brings the ipsilateral ovary and fallopian tube via traction on the attached cranial suspensory ligament, then takes the uterus and contralateral ovary by virtue of the broad ligament [6,18]. This appeared to be the case for this patient. Multiple case reports suggest that elongated ovarian and/or uterine suspensory ligaments contribute to the pathogenesis of indirect inguinal hernia containing uterus, fallopian tubes and bilateral ovaries [4,8]. In contrast, Fowler suggests that the elongation of the suspensory ligament of the ovary may be secondary to traction created by the hernia [13]. These authors advocate for close gynecological follow-up throughout childhood, with concern for increased risk of ovarian torsion [4,8]. There is a known relationship between disorders of sexual development, specifically Müllerian anomalies, and hernia uterine inguinale [4,7,10]. However, at time of surgical correction it is often possible to rule out anatomical abnormalities, and it is unlikely for disorders of sexual development to accompany a case of indirect inguinal hernia containing uterus and bilateral adnexa in an anatomically normal female [7,8]. Discovery of a sliding hernia has conventionally indicated conversion from laparoscopic to open repair, although there have been descriptions of laparoscopic dissection to free the sliding fallopian tube from the hernia sac [19].

3. Conclusion

Inguinal hernias in female infants may contain critical organs including the uterus and bilateral adnexa. Ultrasound is an essential tool for rapid diagnosis and evaluation of organ viability [4–6]. In the absence of strangulation, operation on an urgent rather than emergent basis is appropriate. Initial laparoscopic approach enabled reduction and evaluation of the incarcerated hernia contents as well as evaluation of the contralateral side, with conversion to open repair when the sliding mechanism was noted. Closure of the internal ring was performed to prevent recurrence. The benefits and feasibility of an initial laparoscopic approach warrant consideration in cases of inguinal hernia containing uterus and adnexa.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Funding

No funding or grant support.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.epsc.2019.101330>.

[org/10.1016/j.epsc.2019.101330](https://doi.org/10.1016/j.epsc.2019.101330).

References

- [1] Ziegler MM. Diagnosis of inguinal hernia and hydrocele. *Pediatr Rev* 1994;15: 286–8.
- [2] Kapur P, Caty MG, Glick PL. Pediatric hernias and hydroceles. *Pediatr Clin N Am* 1998;45:773–89.
- [3] Huang CS, Luo CC, Chao HC, et al. The presentation of asymptomatic palpable movable mass in female inguinal hernia. *Eur J Pediatr* 2003;162:493–5.
- [4] George EK, Oudesluys-Murphy AM, Madern GC, et al. Inguinal hernias containing the uterus, Fallopian tube, and ovary in premature female infants. *J Pediatr* 2000; 136(5):696–8.
- [5] Artas H, Gurbuzer N. Inguinal hernia containing both ovaries and the uterus in an infant. *J Ultrasound Med* 2012;31(7):1138–9.
- [6] Derinkuyu BE, Affrancheh MR, Sönmez D, et al. Canal of Nuck hernia in a female infant containing uterus, bilateral adnexa and bowel. *Balkan Med J* 2016;33(5): 566–8.
- [7] Cascini V, Lisi G, Di Renzo D, et al. Irreducible indirect inguinal hernia containing uterus and bilateral adnexa in a premature female infant: report of an exceptional case and review of the literature. *J Pediatr Surg* 2013;48(1):e17–9.
- [8] Karadeniz Cerit K, Ergelen R, Colak E, et al. Inguinal hernia containing uterus, Fallopian tube, and ovary in a premature newborn. *Case Rep Pediatr* 2015;2015: 807309.
- [9] Ming YC, Luo CC, Chao HC, et al. Inguinal hernia containing uterus and uterine adnexa in female infants: report of two cases. *Pediatr Neonatol* 2011;52(2):103–5.
- [10] Akilloğlu I, Kaymakci A, Akkoyun I, et al. Inguinal hernias containing the uterus: a case series of 7 female children. *J Pediatr Surg* 2013;48(10):2157–9.
- [11] Gnidec AA, Marshall DG. Incarcerated direct inguinal hernia containing uterus, both ovaries, and Fallopian tubes. *J Pediatr Surg* 1986;21(11):986.
- [12] Harjai MM. Uncommon content in congenial inguinal hernia. *J Indian Assoc Pediatr Surg* 2014;19(4):244–5.
- [13] Fowler CL. Sliding indirect hernia containing both ovaries. *J Pediatr Surg* 2005;40 (9):e13–4.
- [14] Chang SJ, Chen JY, Hsu CK, et al. The incidence of inguinal hernia and associated risk factors of incarceration in pediatric inguinal hernia: a nation-wide longitudinal population-based study. *Hernia* 2016;20(4):559–63.
- [15] Burgmeier C, Dreyhaupt J, Schier F. Gender-related differences of inguinal hernia and asymptomatic patent processus vaginalis in term and preterm infants. *J Pediatr Surg* 2015;50(3):478–80.
- [16] Nah SA, Giacomello L, Eaton S, et al. Surgical repair of incarcerated inguinal hernia in children: laparoscopic or open? *Eur J Pediatr Surg* 2011;21(1):8–11.
- [17] Juang D, Fraser JD, Holcomb III GW. The laparoscopic approach for repair of indirect inguinal hernias in infants and children. *Transl Pediatr* 2016;5(4):222–6.
- [18] Hutson JM, Kearsley I. Is the ovary in an inguinal hernia 'descended' like a testis or not? *J Pediatr Surg* 2016;51(7):1197–200.
- [19] Ishii T, Yonekura T, Yamauchi K, et al. Laparoscopic repair of sliding inguinal hernia in female children. *Pediatr Surg Int* 2016;32:895–9.