Journal of Pediatric Surgery 55 (2020) 1908–1913

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Other Conditions

Inguinal hernia in girls: A retrospective analysis of over 1000 patients☆

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ARTICLE INFO

Article history:
Received 16 July 2019
Received in revised form 21 February 2020
Accepted 8 March 2020

Key words:
Girls
Inguinal hernia
Hernia repair
Ovary
Strangulation
Androgen insensitivity syndrome

ABSTRACT

Background: In girls with inguinal hernia, timing of surgical repair to prevent ovarian strangulation and screening for Androgen Insensitivity Syndrome (AIS) remain controversial. This study assesses the incidence of ovarian strangulation and AIS, and its associated risk factors.
Methods: Electronic patient records were used to study girls aged 0–15 years who underwent inguinal hernia repair between 2000 and 2017. Patients with incomplete data were excluded. Risk factors were identified using logistic regression.
Results: This study includes 1084 girls (median (IQR) age: 133.5 (14–281) weeks) who underwent 1132 hernia repairs (1015 unilateral, 117 bilateral) within a median (IQR) time interval of 12 (6–23) days following diagnosis. Hernia sac intraoperatively contained ovary in 235 (21.7%) patients, ovary was strangulated in 14 (6%). Risk factors for ovarian strangulation were younger gestational age (OR 0.49), higher birthweight (OR 32.18), and first presentation at the emergency department (OR 13.07). However data were partly missing. Ectopic testis was found in seven (0.6%) patients. Metachronous contralateral inguinal hernia and ipsilateral recurrence developed in 6.1% and 0.3%, respectively.
Conclusions: Ovarian hernia was diagnosed in 21.7%, and ovary was strangulated in 6%. No definite conclusions can be drawn regarding risk factors for strangulation and timing of surgery in girls with irreducible ovarian hernia.
Level of Evidence: Level III.
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The incidence of pediatric inguinal hernia is 0.8–5% and increases to almost 20%% in very low birth weight (<1000 gram) or premature born infants [1,2,32,33]. The risk of incarceration also increases with younger gestational age, and is up to 39% in premature and 3–16% in term infants [3,12]. Other factors associated with increased risk of incarcerated hernia are age less than 1 year and female sex [4]. In girls, the incidence of inguinal hernia is more than four times lower than in boys, however the incidence of bilateral inguinal hernias is almost twice as high as in boys (25.4% vs 12.9%) [2]. Reproductive organs (most commonly ovary) are content of the hernia sac in girls in 15–31% [5], and a small mobile groin tumor is suspect for a herniated ovary, which has a high chance of torsion or strangulation [6,7]. Question remains whether urgent surgical repair is necessary in girls with asymptomatic irreducible hernias containing ovary as the incidence of ovarian strangulation and the rate of oophorectomy following hernia incarceration is not precisely known. Second, there is an ongoing debate about the timing of gonadectomy in case a surgeon finds evidence of Complete or Partial Androgen Insensitivity Syndrome (CAIS/PAIS) in girls who present with inguinal hernia. Since CAIS is characterized by female appearance and normal external female genitals (phenotype) in a genotypically XY male, surgical hernia repair will then unexpectedly reveal testes, while the uterus and ovaries are absent. It has been reported that 90% of the girls with CAIS undergo inguinal hernia repair during childhood [8], and the estimated incidence of CAIS in girls with inguinal hernia is 0.8–2.5% [9–11]. Still, the exact incidence of AIS in our population remains unclear.

In this study, we determined the incidences of ovarian involvement (especially strangulation) and AIS in girls with inguinal hernia, identified risk factors and assessed current treatment strategies in order to formulate clear recommendations on the management of female pediatric inguinal hernia.

☆ Declarations of Interest: None.

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https://doi.org/10.1016/j.jpedsurg.2020.03.015
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1. Material and methods

1.1. Ethical approval

The Medical Ethics Review Committee of the Academic Medical Center Amsterdam reviewed this study (reference number W19_407 # 19.473) and, with a waiver of informed consent requirements, confirmed that the Medical Research Involving Human Subjects Act (WMO) does not apply and an official approval of this study is not required.

1.2. Study setting and data source

In this retrospective cohort study, the medical records of all girls aged 0–15 years who underwent inguinal hernia repair between 1 January 2000 and 30 June 2017 in the Amsterdam University Medical Centers (Emma Children's Hospital AMC and VU University Medical Center) were reviewed. Patients were selected from the Electronic Patient Record using the International Statistical Classification of Diseases and Related Health Problems and the Diagnosis Treatment Combinations codes that are specific for diagnosis of inguinal hernia and its surgical repair. Two authors (KD and RB) independently reviewed all medical records to obtain optimal data collection. Data extraction was completed before 1 November 2017. Appendix A comprises all data that were systematically extracted from the medical records.

1.3. Study population

Female pediatric patients between 0 and 15 years with inguinal hernia who underwent open or laparoscopic inguinal hernia repair were eligible for inclusion. Patients with incomplete or missing operation report were excluded.

1.4. Statistical analysis

Data was assembled and statistical analyses were performed using SPSS software, version 25.0.0.1 (IBM SPSS Statistics). The Kolmogorov–Smirnov test was performed to assess the normality of the distribution of the results. Patient demographics and hernia characteristics are reported as median values with interquartile ranges (IQR) (as the assumption of normality was violated) for continuous variables, and as percentages for categorical variables. Independent samples t-test, χ^2 tests, and Mann–Whitney U tests were used to compare groups. Multivariable logistic regression analysis for strangulation of the ovary was carried out for covariates measurable before inguinal hernia surgery. The investigated factors together with their respective categories were gestational age at birth in weeks, birthweight in kilogram, age at the time of surgery in weeks, location of first presentation (outpatient clinic or emergency department), incarceration and reducibility of the inguinal hernia at physical examination. A backward model building approach was used to construct the multivariable model. Factors investigated for AIS in univariable logistic regression analysis were laterality of the hernia (unilateral or bilateral) and hernia content (unilateral or bilateral) at physical examination. *P* value of <0.05 was considered significant.

2. Results

2.1. Study population

The medical records of 1107 girls were examined for eligibility, 23 patients were excluded because the operation report was incomplete or missing. This cohort includes 1084 patients in whom 1132 open (n = 1129) or laparoscopic (n = 3) hernia repair surgeries were performed. Hernia repair was performed right-sided, left-sided and bilateral in 567 (50.1%), 448 (39.6%) and 117 (10.3%) cases, respectively. Median (IQR) age at the time of surgery was 133.5 (14–281) weeks (Table 1A).

2.2. Clinical findings at first presentation

Diagnosis of inguinal hernia was based on the child's medical history and physical examination, i.e. inspection and palpation of the groin(s). The majority of inguinal hernias was diagnosed at the outpatient clinic in 72.8% (n = 824). In 53 cases (4.7%), inguinal hernia was diagnosed at the emergency department, in 5 (0.4%) during hospital admission, and location of first presentation and subsequent diagnosis of inguinal hernia was unknown in 250 (22.1%) cases (Table 1A). During physical examination by pediatric surgeons, inguinal hernia was reported to be incarcerated in 12 (1%) cases. Ovary was thought to be present in the sac of 163 (14.4%) hernias, of which in seven bilateral. No signs of ovarian herniation were reported in 712 (62.9%) hernias and in 257 (22.7%) cases these data were not available.

2.3. Surgical findings

Median (IQR) time from diagnosis to surgery was 30 (14–60) days. Contralateral groin exploration at the time of unilateral hernia repair was performed in 164 (16.2%) patients. Upon prophylactic exploration the processus vaginalis was found to be open, and was subsequently closed in the same session in 64.6% of these patients. Metachronous contralateral inguinal hernia (MCIH) developed postoperatively in 52/851 (6.1%) patients who underwent unilateral hernia repair without contralateral exploration. Three patients (0.3%) presented with ipsilateral recurrent inguinal hernia (Table 1B).

2.3.1. Ovary as content of the hernia sac

During surgery, ovary was found as content of the hernia sac in 235/1084 (21.7%) patients (Table 1B). In 97 hernias (41.3%), herniated ovary

Table 1A
Baseline characteristics of 1084 patients and 1132 inguinal hernias.

Patient characteristics (n = 1084 patients)	
Female	1084 (100)
Age at surgery, median (IQR), weeks	133.5 (14–281)
Birth status	
Premature (< 37 weeks gestation)	221 (20.4)
Term (37–42 weeks gestation)	297 (27.4)
Serotine (≥ 42 weeks gestation	6 (0.6)
Missing	*560 (51.7)*
Birth weight, median (IQR), gram	2320 (1415–3060)
Missing	*721 (66.5)*
Hernia characteristics (n = 1132 inguinal hernias)	
Initial hernia side	
Right-sided	567 (50.1)
Left-sided	448 (39.6)
Bilateral	117 (10.3)
Diagnosis of inguinal hernia	
Outpatient clinic	824 (72.8)
Emergency department	53 (4.7)
Hospital admission	5 (0.4)
Missing	*250 (22.1)*
Reducibility at physical examination	
Reducible hernia	87 (7.7)
Irreducible hernia	101 (8.9)
Missing	*944 (83.4)*
Incarceration at physical examination	
Yes	12 (1)
No	183 (16.2)
Missing	*937 (82.8)*
Ovary in hernia sac at physical examination[a]	
No	712 (62.9)
Yes, unilateral	156 (13.8)
Yes, bilateral	7 (0.6)
Missing	*257 (22.7)*

Data presented as median (IQR) for continuous variables and n (%) for categorical variables, unless stated otherwise. IQR, Interquartile range.
[a] During physical examination, hernia sac was suspected to contain ovary.

Table 1B
Surgical findings in 1132 hernia repair surgeries.

Pre- and perioperative	
Time from diagnosis to surgery, median (IQR), days	30 (14–60)
Missing	*254 (22.4)*
Urgency	
Elective surgery	1087 (96)
Urgent (<48 hours)	21 (1.9)
Emergency surgery (<24 hours)	24 (2.1)
Ovary in hernia sac	
Yes	235 (20.8)
No	897 (79.2)
Contralateral exploration[a]	
Yes	164 (16.2)
No	851 (83.8)
Postoperative	
Development of MCIH[b]	
Yes	52 (6.1)
No	799 (93.9)
Complications	
Ipsilateral recurrence	3 (0.3)

Data presented as median (IQR) for continuous variables and n (%) for categorical variables, unless stated otherwise. MCIH, metachronous contralateral inguinal hernia; IQR, Interquartile range.
[a] Not applicable to 117 patients who presented with bilateral inguinal hernia.
[b] Not applicable to 117 patients who presented with bilateral inguinal hernia and 164 patients who underwent contralateral exploration.

was accompanied by the fallopian tube. Physical examination at first presentation accurately predicted the intraoperative presence of ovary in 108/163 (66.3%) patients. False negative rate of physical examination was 10.1%, as 72/712 patients had ovary in the hernia sac during surgery without clinical signs of ovarian herniation during physical examination. Median (IQR) time interval between diagnosis and surgery in 163 patients with suspected ovarian hernia was 11 days (6–18).

In patients with ovary in the hernia sac during surgery, the hernia was reported to be irreducible during physical examination at first presentation in 101/235 (43%) patients, reducible in 87/235 (37%) patients, and in 47/235 (20%) patients data regarding reducibility of the inguinal hernia was missing. Girls with herniated ovary during surgery were younger (12 vs 197 weeks), had a lower birth weight (2190 vs 2433 gram) and were operated earlier after diagnosis (12 vs 37 days) than girls without ovary as content of the hernia sac (Table 2). Gestational age at birth was not different between the groups (p = 0.05).

2.3.2. Ovarian strangulation

In 14/235 (6.0%) patients, ovary was intraoperatively strangulated. The hernia was described to be irreducible during physical examination at first presentation in 10/14 (71.4%) patients (data from one patient was missing). Median (IQR) age at the time of surgery was 13 (5.8–22) weeks, the median (IQR) time interval between diagnosis and surgery was 11.5 (1.3–20.5) days, ranging from 0 to 49 days (Table 2). Hernia repair was performed within a maximum of 14 days after diagnosis was made at the emergency department or 49 days after visiting the outpatient clinic for diagnosis of inguinal hernia. Three patients underwent an emergency operation and were operated within 24 hours after diagnosis. In 13 patients, ovary recovered after intraoperative repositioning and, if necessary, detorsion. There are no long-term follow-up data on the postoperative vitality after repositioning of the strangulated ovaries. During surgery in one patient who presented with an incarcerated, irreducible hernia in 2007, the ovary was completely necrotic and did not show any vitality. Therefore a salpingo-oophorectomy was performed.

Gestational age at birth, birth weight, age at surgery, initial hernia side and waiting time for surgery were not different between patients with and without ovarian strangulation (Table 2).

2.4. Risk factors for ovarian strangulation in patients with ovarian hernias

Data from 235 patients with intraoperatively ovary in the hernia sac were included in the risk analysis for ovarian strangulation (Table 3). The risk of ovarian strangulation in irreducible ovarian hernias is 9.9% compared to a 3.4% risk in reducible ovarian hernias. Multivariable regression analysis using backward selection showed that younger gestational age at birth (OR 0.49, 95% CI 0.28–0.86, p = 0.01), higher birth weight (OR 32.18, 95% CI 2.05–506.01, p = 0.01) and first presentation at the emergency department (OR 13.07, 95% CI 1.93–88.51, p = 0.008) were significantly associated with the odds of ovarian strangulation. Hernia side, age at the time of surgery, incarceration and reducibility of the hernia during physical examination were not independently associated with ovarian strangulation.

Table 2
Patient demographics and hernia characteristics in (I) Patients (n = 1084) with and without ovary as content of the hernia sac during hernia repair surgery (n = 1132) and (II) Patients (n = 235) with ovary as content of the hernia sac during hernia repair.

	I			II		
	Ovary n = 235	No ovary n = 897	*p* value	Strangulated n = 14	Vital ovary n = 221	*p* value
Gestational age at birth						
<37 weeks gestation	88 (37.4)	143 (15.9)	0.05	5 (35.7)	83 (37.6)	0.87
≥37 weeks gestation	96 (40.9)	223 (24.9)		6 (42.9)	90 (40.7)	
Missing	*51 (21.7)*	*531 (59.2)*		*3 (21.4)*	*48 (21.7)*	
Birth weight, median (IQR), gram	2190 (1215–2900)	2433 (1548–3203)	0.004	2400 (750–3120)	2175 (1261–2895)	0.97
Missing	*88 (37.4)*	*663 (73.9)*		*7 (50)*	*81 (36.7)*	
Presentation						
Outpatient clinic	144 (61.3)	680 (75.8)	< 0.001	6 (42.9)	138 (62.4)	0.007
Emergency department	25 (10.6)	28 (3.1)		6 (42.9)	19 (8.6)	
Hospital admission	3 (1.3)	2 (0.2)		0 (0)	3 (1.4)	
Missing	*63 (26.8)*	*187 (20.9)*		*2 (14.3)*	*61 (27.6)*	
Age at hernia operation, median (IQR), weeks	12 (8–18)	197 (43–305)	<0.001	13 (5.8–22)	12 (8–17.5)	0.91
Initial hernia side						
Right-sided	96 (40.9)	471 (52.5)	< 0.001	6 (42.9)	90 (40.7)	0.98
Left-sided	120 (51)	328 (36.6)		7 (50)	113 (51.1)	
Bilateral	19 (8.1)	98 (10.9)		1 (7.1)[a]	18 (8.2)[b]	
Time between diagnosis and operation, median (IQR), days	12 (6–23)	37 (18–67)	<0.001	11.5 (1.3–20.5)	12.5 (6.3–23)	0.19
Missing	*63 (26.8)*	*191 (21.3)*		*2 (14.3)*	*61 (27.6)*	

Data presented as n (%), unless stated otherwise. IQR, Interquartile range.
[a] Right-sided ovary was strangulated.
[b] Left-sided ovary was strangulated in 11 patients, right-sided ovary in seven patients.

Table 3
Adjusted results from logistic regression analysis of covariates for ovarian strangulation in 235 girls with ovarian hernia.

	Strangulation rate n (%)	Odds ratio (95% CI)	*p* value
Gestational age at birth, weeks' gestation	11 of 184 (6)	0.49 (0.28, 0.86)	0.01
Birth weight, kilogram	7 of 147 (4.8)	32.18 (2.05, 506.01)	0.01
Age at hernia operation, weeks	14 of 235 (6)		
Presentation			
Outpatient clinic	6 of 146 (4.1)		
Emergency department	6 of 26 (23.1)	13.07 (1.93, 88.51)	0.008
Hernia side			
Right-sided	7 of 104 (6.7)		
Left-sided	7 of 131 (5.3)		
Hernia at physical examination			
Not incarcerated	8 of 183 (4.4)		
Incarcerated	5 of 12 (41.7)		
Hernia at physical examination			
Not reducible	10 of 101 (9.9)		
Reducible	3 of 87 (3.4)		

CI, confidence interval.

2.5. Androgen insensitivity syndrome (AIS)

During surgery, testes were present in the hernia sac in seven (0.6%) patients (median (IQR) age at time of surgery: 17 (5–21) weeks). At preoperative physical examination one (28.6%) or two (71.4%) gonads were palpable. In six patients gonadectomy was performed during (n = 4) or after (n = 2) hernia repair at a median (IQR) age of 20.5 (13.5–33.5) weeks. Postoperative pathological examination showed normal testicular tissue without any malignant changes in all removed gonads.

2.6. Risk factors for AIS

In univariable analysis, patients who presented with bilateral inguinal hernia were at largely increased risk for AIS (OR 22.61, 95% CI 4.34–117.90, p < 0.001). The risk increased to almost 200-fold if during preoperative physical examination there was bilateral hernia content (OR 192.50, 95% CI 22.36–1657.28, p < 0.001) (Table 4).

3. Discussion

This retrospective cohort study including 1084 girls with inguinal hernia showed that the incidence of ovary as content of the hernia sac in girls with inguinal hernia is 21.7%, 101/1084 (9.3%) girls had irreducible ovarian hernias. Ovary was perioperatively strangulated in 6% of the girls with ovarian hernias.

The incidence of ovarian herniation and ovarian strangulation corresponds well to previously described incidences that were reported to be 15–31% and 2–33%, respectively [5,12,13]. The large variability regarding the incidence of ovarian strangulation is probably caused by the use of different definitions in various studies: some included all patients with irreducible hernias, while others only included patients with incarcerated hernias, i.e. symptomatic irreducible hernias. Previously reported incidences of irreducible ovarian hernias in girls also largely differ. Boley et al. and Takehara et al. found an incidence of irreducible ovary in girls with inguinal hernia of 4% and 5%, respectively, whereas Rowe et al. demonstrated that 29% of 259 girls had irreducible inguinal hernias [6,7,14]. Corresponding to our results, Hirabayashi et al. more recently studied 673 girls and found an incidence of 10.8% [15].

Younger gestational age, lower birth weight and first presentation at the emergency department could be considered as potential risk factors for ovarian strangulation, though no firm conclusions can yet be drawn as a considerable amount of data from this cohort was missing. The risk of ovarian strangulation in irreducible ovarian hernias is 9.9% compared to a 3.4% risk in reducible ovarian hernias. However, on multivariable analysis in our cohort, incarceration and reducibility of the inguinal hernia were not found to be associated with an increased risk for strangulation. We assume that asymptomatic irreducible ovaries do not seem at a large risk for compression of their blood supply, although alteration of the normal anatomy when ovary slides into the hernia sac makes torsion more likely [6]. Chen et al. recently demonstrated that larger ovaries (volume ≥ 5 cm^3) were more likely to result in ovarian torsion in 32 female infants. As ovarian volume declines with increasing age, ovarian hernias in female patients younger than one year old might be more likely to be incarcerated and twisted, subsequently leading to strangulation of the ovary [16]. In addition, recent results of Lee et al. indicated that in female pediatric patients (n = 1210) younger age is associated with a higher risk for incarceration of the ovary [17]. This corresponds to the results of our study, since in our cohort ovarian strangulation only occurred in patients that were aged between 4 and 54 weeks old.

In our cohort, ovary could be preserved in 234 patients (99.6%) if hernia repair was performed within a median (IQR) of 12 (6–23) days. This time interval was longer than advised by many surgeons [6,18–20], however, the waiting time did not affect the extirpation rate of a strangulated ovary: only one salpingo-oophorectomy was performed within 24 hours after detection of the incarcerated hernia. Unfortunately, follow-up data on the postoperative viability after repositioning of the ovaries were not available due to the retrospective study design. Over the past decade, it is widely accepted to leave a previously strangulated ovary in situ to permit survival. However, surgery in this patient took place almost 13 years ago. Esposito et al. operated on 16 girls with asymptomatic irreducible inguinal hernias containing prolapsed ovaries between one to four days after diagnosis and reported no ovarian complications, whereas Hirabayashi et al. followed their policy to postpone surgical repair in girls until after nine months of age, and also reported no cases of ovarian torsion [15,20]. Zamakhshary et al. previously found an almost 2-fold risk of incarcerated hernia (OR 1.92, 95% CI 1.11–3.32) [4] in children below 2 years of age if hernia repair was performed more than 14 days after diagnosis. In contrast to irreducible incarcerated inguinal hernias that should be treated as an emergency [17], we endorse performing surgical repair in girls with reducible and asymptomatic irreducible ovarian hernias shortly after diagnosis. Our institution therefore retains the policy to try to perform surgical repair in girls with suspected ovarian hernia within two weeks after diagnosis.

Table 4
Unadjusted results of logistic regression analysis of covariates for androgen insensitivity syndrome in 1084 girls with 1132 inguinal hernias.

	Androgen insensitivity syndrome n (%)	Odds ratio (95% CI)	*p* Value
Laterality of inguinal hernia			
Unilateral (i.e. left or right-sided)	2 of 1015 (0.2)	1 [Reference]	
Bilateral	5 of 117 (4.3)	22.61 (4.34, 117.90)	<0.001
Hernia content at preoperative physical examination [a]			
Unilateral content	2 of 156 (1.3)	1 [Reference]	
Bilateral content	5 of 7 (71.4)	192.50 (22.36, 1657.28)	<0.001

CI, confidence interval.
[a] In 712 patients, there was no hernia content during preoperative physical examination, the results of 257 patients were missing.

Current literature and guidelines recommend detorsion and subsequent ovarian preservation after torsion instead of oophorectomy [21–23]. Parelkar et al. showed that 12 out of 13 (92%) children with ovarian torsion, of which the majority was intraoperatively graded as moderately to severely ischemic/necrotic, showed good vascularity and follicular development 3 months after detorsion [24]. Esposito et al. also showed morphologically normal ovaries with good vascular supply at postoperative follow-up in their 16 patients with asymptomatic irreducible ovarian inguinal hernias [20]. Moreover, there is no clear evidence of thromboembolic events after detorsion, and oophoropexy is not necessarily indicated after ovarian torsion [23]. We therefore believe that oophorectomy should not be performed after detorsion and repositioning of a strangulated ovary in case it shows any signs of revitalization.

Limitations of this study were mostly attributed to the retrospective study design and include that some of the retrieved data were incomplete and thus not available. Consequently, the exact time of occurrence of the inguinal hernia is unknown.

CAIS occurs in phenotypical female patients in whom secretion of antimullerian hormone by Sertoli cells of the testes results in absence of the uterus, cervix and proximal vagina. The presence of gonads in the hernia sac, bilateral hernias or a family history of CAIS is known to trigger consideration of CAIS diagnosis [25]. In our cohort, presentation with bilateral inguinal hernia (OR 22.61, $p < 0.001$) and bilateral presence of gonads (OR 192.50, $p < 0.001$) were indeed identified as risk factors for AIS. Analysis of CAIS in 120 patients by Deeb et al. revealed that 57% presented with inguinal hernia among which the incidence of unilateral and bilateral hernia was equally dispersed and only a third of the patients had palpable gonads [25].

Controversy still exists in children with AIS to perform gonadectomy in combination with surgical hernia repair to minimize the risk of gonadal malignancy, or whether gonadectomy is better delayed to allow for spontaneous induction of puberty. In this cohort, six patients underwent gonadectomy at a median (IQR) age of 20.5 (13.5–33.5) weeks. The latest gonadectomy was performed more than eight years ago and pathological examination showed no malignant changes in any of the testes.

Prevalence of malignant germ cell tumors was thought to be 22% in children with AIS [26], although more recent data shows that the risk of premalignant or malignant change in germ cells is low before and during puberty with an estimated germ cell tumor prevalence of 0.8% [27,28]. In addition, no malignant changes were observed in the gonads of 14 patients who underwent gonadectomy at an average age of 18.2 years (range 2–21 years) and only once before completing puberty [29]. Gonadectomy should therefore be deferred until after puberty since it does not substantially increase the risk of malignancy, however it does enable development of secondary sex characteristics and avoids early hormone replacement [28,30]. Preservation of at least one testicular gonad until puberty is recommended to preserve self-esteem in adolescents who spontaneously achieve puberty and to enable optimal breast development and improved bone density [31]. It is therefore not necessary that the surgeon who unexpectedly encounters testicular gonads during female pediatric hernia repair immediately has to decide on any additional treatment e.g. removal of the gonads. After completion of hernia repair surgery, the patient suspected of AIS can be referred to an appropriate specialist for diagnostic workup and further treatment.

4. Conclusion

Ovarian inguinal hernia was diagnosed in 21.7% of the patients, and ovary was strangulated in 6%. Still, it is hard to accurately predict whether there is ovary or a strangulated ovary present in the hernia sac during surgery by clinical preoperative assessment. No definite conclusions can be drawn regarding risk factors for strangulation and timing of surgery in girls with irreducible ovarian hernia.

Acknowledgement

The authors would like to thank N.A.G. Hakkenbrak for her contributions regarding the selection of patients and data acquisition.

Funding

This research is funded by The Netherlands Organization for Health Research and Development (ZonMw), project number 852001903.

Appendix A. Systematically extracted data from the medical patient records

- Patient characteristics, i.e.:
  - o Gestational age at birth (premature <37 weeks, full-term ≥37 weeks)
  - o Birth weight
  - o Medical history and comorbidities
  - o Age at time of surgery
- Hernia characteristics, i.e.:
  - o First presentation (e.g. at outpatient clinic or emergency department)
  - o Side of inguinal hernia
  - o Physical examination of the groin(s)
- Surgical findings, i.e.:
  - o Urgency of procedure (elective vs. emergency surgery)
  - o Hernia repair technique (open or laparoscopic inguinal hernia repair)
  - o Content of the hernia sac
  - o Ovarian strangulation
  - o Appearance of internal reproductive organs
  - o Contralateral exploration
- Postoperative findings, i.e.:
  - o Pathology reports
  - o Postoperative complications (recurrence)
  - o Metachronous contralateral inguinal hernia

References

[1] Burgmeier C, Dreyhaupt J, Schier F. Comparison of inguinal hernia and asymptomatic patent processus vaginalis in term and preterm infants. J Pediatr Surg 2014; 49:1416–8. https://doi.org/10.1016/j.jpedsurg.2014.03.013.
[2] Chang SJ, Chen JYC, 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;20:559–63. https://doi.org/10.1007/s10029-015-1450-x.
[3] Wang KS, Committee on F, Newborn AA of P, Section on Surgery AA of P. Assessment and management of inguinal hernia in infants. Pediatrics 2012;130:768–73. https://doi.org/10.1542/peds.2012-2008.
[4] Zamakhshary M, To T, Guan J, et al. Risk of incarceration of inguinal hernia among infants and young children awaiting elective surgery. CMAJ 2008;179:1001–5. https://doi.org/10.1503/cmaj.070923.
[5] 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:e17–9. https://doi.org/10.1016/j.jpedsurg.2012.09.065.
[6] Boley SJ, Cahn D, Lauer T, et al. The irreducible ovary: a true emergency. J Pediatr Surg 1991;26:1035–8. https://doi.org/10.1016/0022-3468(91)90668-j.
[7] Takehara H, Hanaoka J, Arakawa Y. Laparoscopic strategy for inguinal ovarian hernias in children: when to operate for irreducible ovary. J Laparoendosc Adv Surg Tech A 2009;19(Suppl. 1):S129–31. https://doi.org/10.1089/lap.2008.0204.supp.
[8] Viner RM, Teoh Y, Williams DM, et al. Androgen insensitivity syndrome: a survey of diagnostic procedures and management in the UK. Arch Dis Child 1997;77:305–9. https://doi.org/10.1136/adc.77.4.305.
[9] Gans SL, Rubin CL. Apparent female infants with hernias and testes. Am J Dis Child 1962;104:82–6. https://doi.org/10.1001/archpedi.1962.02080030084012.
[10] Jagiello G, Atwell JD. Prevalence of testicular feminisation. Lancet 1962;279:329. https://doi.org/10.1016/S0140-6736(62)91289-8.
[11] Sarpel U, Palmer SK, Dolgin SE. The incidence of complete androgen insensitivity in girls with inguinal hernias and assessment of screening by vaginal length measurement. J Pediatr Surg 2005;40:133–7. https://doi.org/10.1016/j.jpedsurg.2004.09.012.
[12] Ein SH, Njere I, Ein A. Six thousand three hundred sixty-one pediatric inguinal hernias: a 35-year review. J Pediatr Surg 2006;41:980–6. https://doi.org/10.1016/J.JPEDSURG.2006.01.020.

[13] Erdoğan D, Karaman İ, Aslan MK, et al. Analysis of 3776 pediatric inguinal hernia and hydrocele cases in a tertiary center. J Pediatr Surg 2013;48:1767–72. https://doi.org/10.1016/J.JPEDSURG.2012.09.048.
[14] Rowe MI, Clatworthy HW. Incarcerated and strangulated hernias in children. A statistical study of high-risk factors. Arch Surg 1970;101:136–9. https://doi.org/10.1001/archsurg.1970.01340260040006.
[15] Hirabayashi T, Ueno S, Hirakawa H, et al. Surgical treatment of inguinal hernia with prolapsed ovary in young girls: emergency surgery or elective surgery. Tokai J Exp Clin Med 2017;42:89–95.
[16] Chen Y, Peng X-Z, Lu W, et al. Risk factors for strangulated ovarian hernia in female infants: the role of ovarian volume. Curr Med Sci 2018;38:1032–7. https://doi.org/10.1007/s11596-018-1980-4.
[17] Lee SR. Efficacy of laparoscopic herniorrhaphy for treating incarcerated pediatric inguinal hernia. Hernia 2018;22:671–9. https://doi.org/10.1007/s10029-017-1655-2.
[18] Houben CH, Chan KWE, Mou JWC, et al. Irreducible inguinal hernia in children: how serious is it? J Pediatr Surg 2015;50:1174–6. https://doi.org/10.1016/j.jpedsurg.2014.10.018.
[19] El Gohary A. Sliding ovary inside a hernia sac; a true emergency q. J Pediatr Surg Case Reports 2014;2:522–3. https://doi.org/10.1016/j.epsc.2014.10.001.
[20] Esposito C, Gargiulo F, Farina A, et al. Laparoscopic treatment of inguinal ovarian hernia in female infants and children: standardizing the technique. J Laparoendosc Adv Surg Tech 2019;29:568–72. https://doi.org/10.1089/lap.2018.0630.
[21] Campbell BT, Austin DM, Kahn O, et al. Current trends in the surgical treatment of pediatric ovarian torsion: we can do better. J Pediatr Surg 2015;50:1374–7. https://doi.org/10.1016/j.jpedsurg.2015.04.018.
[22] Geimanaite L, Trainavicius K. Ovarian torsion in children: management and outcomes. J Pediatr Surg 2013;48:1946–53. https://doi.org/10.1016/j.jpedsurg.2013.04.026.
[23] Dasgupta R, Renaud E, Goldin AB, et al. Ovarian torsion in pediatric and adolescent patients: a systematic review. J Pediatr Surg 2018;53(7):1387–91.
[24] Parelkar SV, Mundada D, Sanghvi BV, et al. Should the ovary always be conserved in torsion? A tertiary care institute experience. J Pediatr Surg 2014;49:465–8. https://doi.org/10.1016/j.jpedsurg.2013.11.055.
[25] Deeb A, Hughes IA. Inguinal hernia in female infants: a cue to check the sex chromosomes? BJU Int 2005;96:401–3. https://doi.org/10.1111/j.1464-410X.2005.05639.x.
[26] Morris JM. The syndrome of testicular feminization in male pseudohermaphrodites. Am J Obstet Gynecol 1953;65:1192–211. https://doi.org/10.1016/0002-9378(53)90359-7.
[27] Cools M, Drop SLS, Wolffenbuttel KP, et al. Germ cell tumors in the intersex gonad: old paths, new directions, moving frontiers. Endocr Rev 2006;27:468–84. https://doi.org/10.1210/er.2006-0005.
[28] Purves JT, Miles-Thomas J, Migeon C, et al. Complete androgen insensitivity: the role of the surgeon. J Urol 2008;180:1716–9. https://doi.org/10.1016/j.juro.2008.03.110.
[29] Hannema SE, Scott IS, Rajpert-De Meyts E, et al. Testicular development in the complete androgen insensitivity syndrome. J Pathol 2006;208:518–27. https://doi.org/10.1002/path.1890.
[30] Patel V, Casey RK, Gomez-Lobo V. Timing of gonadectomy in patients with complete androgen insensitivity syndrome–current recommendations and future directions. J Pediatr Adolesc Gynecol 2016;29:320–5. https://doi.org/10.1016/j.jpag.2015.03.011.
[31] Cheikhelard A, Morel Y, Thibaud E, et al. Long-term follow up and comparison between genotype and phenotype in 29 cases of complete androgen insensitivity syndrome. J Urol 2008;180:1496–501. https://doi.org/10.1016/j.juro.2008.06.045.
[32] Rajput A, Gauderer MWL, Hack M. Inguinal hernias in very low birth weight infants: Incidence and timing of repair. J Pediatr Surg 1992;27(10):1322–4. https://doi.org/10.1016/0022-3468(92)90287-H.
[33] Kumar VHS, Clive J, Rosenkrantz TS, et al. Inguinal hernia in preterm infants (≤32-Week Gestation). Pediatr Surg Int 2002;18(2–3):147–52. https://doi.org/10.1007/s003830100631.