

A Rare Pathology Mimicking an Irreducible Inguinal Hernia in Girls: Cyst in the Canal of Nuck

Mehmet Arpacık¹, Aytekin Kaymakçı¹, Turan Yıldız², Ceyhan Şahin¹, Hayriye Nihan Ayyıldız Karaman¹, Zeliha Akış Yıldız¹, Semih Lütfi Mirapoglu¹, Fatma Tuğba Güvenç¹, İtir Ebru Zemheri³

¹Department of Pediatric Surgery, University of Health Sciences Umraniye Training and Research Hospital, Istanbul, Turkey

²Department of Pediatric Surgery, Inonu University, Turgut Ozal Medical Center, Malatya, Turkey

³Department of Pathology, University of Health Sciences Umraniye Training and Research Hospital, Istanbul, Turkey

Abstract

Introduction: Cyst in the canal of nuck (CCN) is a result of a closure defect during the processus vaginalis in girls. The symptoms and mass findings in the inguinal region can mimic an irreducible inguinal hernia. The aim of this study was to investigate the differential diagnosis and surgical findings of girls who were operated on for an irreducible inguinal hernia and the frequency of consideration of a CCN.

Methods: The data of 868 female patients aged 0–16 years who were referred to surgery with the preliminary diagnosis of an inguinal hernia between 2004 and 2019 were analyzed retrospectively.

Results: A total of 138 patients (15.89%) were sent to emergency surgery for an inguinal hernia that could not be reduced. In 15 (10.8%) of these patients, the hernia spontaneously reduced after the application of general anesthesia. In the remaining patients, in order of decreasing frequency, the findings were ovarian sliding hernia (n=93; 67.3%), CCN (n=17; 12.3%), herniated small intestine (n=12; 8.6%), and lymphadenitis (n=1; 0.07%). The mean age of the patients diagnosed with CCN (52.13 months) was significantly higher than the mean age of patients with an incarcerated inguinal hernia (7.23 months).

Discussion and Conclusion: CCN is seen more frequently than expected. Especially, the differential diagnosis should be made with irreducible inguinal hernia in older than 2 years.

Keywords: Cyst in the canal of nuck; girls; irreducible inguinal hernia.

The canal of nuck is formed in girls as a result of peritoneal invagination that extends into the inguinal canal with the round ligament. It is analogous to a patent processus vaginalis in males. An indirect inguinal hernia or cyst can develop due to the insufficient closure. If the defect is large, herniation of the small intestine, colon, omentum, ovaries/Fallopian tubes, or uterus may occur. An indirect inguinal

hernia manifests with swelling in the groin. Incarceration of irreducible tissue in the hernia sac may cause circulatory disorders. A cyst in the canal of nuck (CCN) is similar to an indirect inguinal hernia in that it is painless and not reducible into the groin; however, only fluid is trapped within the hernia sac. Inguinal lymphadenopathy, lipoma, and tumors located in this region may have similar clinical findings^[1-3].

Correspondence (İletişim): Mehmet Arpacık, M.D. Sağlık Bilimleri Üniversitesi Umraniye Eğitim Araştırma Hastanesi, Çocuk Cerrahi Bölümü, İstanbul, Turkey

Phone (Telefon): +90 505 617 83 05 **E-mail (E-posta):** mehmetarpacik@hotmail.com

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Most surgeons find a clinical and physical examination to be sufficient for the diagnosis of inguinal hernia. Sometimes ultrasonography (US) is used to confirm the diagnosis. In complicated cases, computed tomography (CT) or magnetic resonance imaging (MRI) may also be used. However, the preoperative diagnosis and the postoperative surgical diagnosis may differ^[4]. The treatment for patients with a diagnosis of incarcerated irreducible inguinal hernia is emergency surgery. Cases of hydrocele in males are generally left undisturbed until the age of 2 years due to the possibility of natural regression^[3]. However, there is no similar guidance in the literature about the timing of surgical treatment in cases of CCN, which is the counterpart to hydrocele in males.

The objective of this study was to evaluate a large series of hernias diagnosed in girls and discuss the surgical results of these patients who underwent urgent surgery due to an irreducible inguinal hernia, and to draw attention to CCN, which mimics an irreducible inguinal hernia. An analysis was conducted of the complaints, examination findings, preliminary diagnoses, surgical findings, histopathological diagnoses, and surgical results of female patients who were operated on due to inguinal hernia by pediatric surgeons in three centers during a 16-year period.

Materials and Methods

On receipt of approval from the local Ethics Committee (11337-05.31.2019), a retrospective search of hospital information systems was performed to collect the data of female patients referred to surgery with the initial diagnosis of inguinal hernia in the pediatric surgery clinics of the Health Sciences University Umraniye Training and Research Hospital, İnönü University Faculty of Medicine Hospital, and Sivas Numune Hospital.

The diagnosis of the patients was made based on a clinical and physical examination. As part of the differential diagnosis, 34 patients underwent a US examination. The same pediatric surgeons performed the diagnosis of inguinal hernia and surgical treatment. Reduction was attempted in all patients who were considered to have

an incarcerated inguinal hernia, and those with an irreducible hernia were taken to emergency surgery. Open surgery was used to apply high ligation to the hernia sac through a standard transverse inguinal skin crease incision and the external ring was tightened. Patients with a hernia that could be reduced preoperatively and those with a hernia that spontaneously reduced prior to surgery were excluded from the study. A total of 138 patients with a preliminary diagnosis of emergency irreducible inguinal hernia were evaluated.

The cases were grouped as right, left, and bilateral hernias according to the location of the pathology. The contents of the hernia sac and its anatomical structure were analyzed to determine if the findings were a sliding hernia, CCN, intestine material, or lymphadenopathy. Postoperative complications, histopathological results, and recurrence were assessed.

Statistical Analysis

IBM SPSS Statistics for Windows, Version 22.0 (IBM Corp., Armonk, NY, USA) was used to examine the study data. Normal distribution was evaluated with the Shapiro-Wilks test and it was found that age did not show normal distribution. Descriptive statistics (mean, SD, median) were used to analyze the data, and the Mann-Whitney U test was applied to compare the parameter of age between the two groups. A $p < 0.05$ was considered significant.

Results

The study group comprised 138 children, with an age range from 6 days to 187 months, with a mean age of 12.76 ± 28.76 months and a median age of 1.8 months. Those with a diagnosed irreducible inguinal hernia were aged between 6 days and 82.5 months, with a mean age of 7.23 ± 15.45 months and a median age of 1.7 months. The age of patients with CCN ranged from 2 months to 187 months, with a mean age of 52.13 ± 58.41 months and a median age of 30 months. The mean age of the CCN patients was statistically significantly higher than that of the irreducible inguinal hernia group ($p = 0.000$; $p < 0.05$) (Table 1).

Table 1. Evaluation of the groups according to age

Group	Minimum	Maximum	Mean±SD	Median	p
Incarcerated hernia (n=121)	6 days	82.5 months	7.23±15.45	1.7 months	0.000*
Nuck canal cyst (n=17)	2 months	187 months	52.13±58.41	30 months	
Total	6 days	187 months	12.76±28.76	1.8 months	

Mann-Whitney U test. * $p < 0.05$.

Among the patients diagnosed with irreducible hernias who underwent emergency surgery, 77 had swelling on the right side of the groin, 48 on the left, and 13 on both sides. The US was used in the differential diagnosis in 34 cases. US findings were evaluated as an ovarian sliding hernia in 28 patients, a segment of the intestine in five patients, and a cystic mass in one patient. A 4-month-old patient who was diagnosed with CCN based on the US results was followed up without surgery in the short term (Fig. 1).

In the five patients with US findings of intestinal hernia content, 2 cases were not confirmed during surgery because the hernia was reduced after the administration of general anesthesia, and in 3 cases the hernia sac was found to contain a segment of the intestine. An ovarian sliding hernia was reduced after the administration of general anesthesia in 3 of 28 patients. Two cases reported as ovary/ovarian cyst in the hernia sac based on US imaging was found to be CCN. The US and surgical diagnoses of 23 patients were compatible with a sliding hernia; however, 4 patients had an ovarian cyst or multiple cysts.

In 15 of 138 cases of female patients who were sent for emergency surgery because the hernia could not be reduced, spontaneous reduction was observed after the administration of general anesthesia. The surgical results were

right ovarian sliding hernia (n = 50), right CCN (n = 11), right inguinal hernia (n=7.5 containing intestinal material, 2 of intestines and the appendix), right inguinal lymphadenitis (n=1), left ovarian sliding hernia (n=35, 2 containing ovaries and uterus), left CCN (n=5), left inguinal hernia with intestinal contents (n=3), bilateral ovarian sliding hernia (n=8), bilateral inguinal hernia with intestinal contents (n=2), and bilateral CCN (n=1) (Table 2).

Resection was not required in any of the cases of a hernia sac with intestinal content because blood circulation of the bowels was satisfactory. Ovarian torsion was observed in two patients; in one case, the circulation of the ovary improved when the ovary was detorsioned, while in the other, an oophorectomy was performed because necrosis had developed. In the patient with lymphadenopathy, the lymph node was excised and histopathologically reported as a reactive lymph node. Seventeen patients had CCN (Fig. 2). The cysts were excised by ligating the remnants of the processus vaginalis. Histopathologically, the wall of the CCN was partially lined with stratified squamous, transitional and columnar epithelium, and contained moderately chronic active inflammation in focal areas (Fig. 3). No recurrence was observed during the postoperative follow-up 12 months of the study patients.

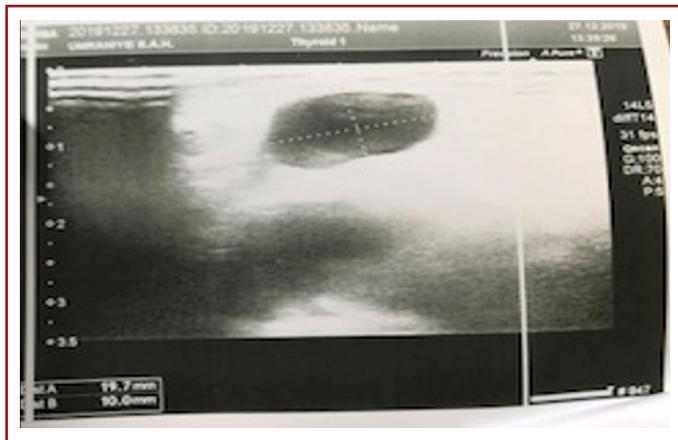


Figure 1. Ultrasonography image of a 20×10 mm non-abdominal cystic mass located in the subcutaneous tissue of the left inguinal region.



Figure 2. Nuck canal cyst in the left inguinal region.

Table 2. Surgical outcomes of the irreducible hernias

Diagnosis	Right	Left	Bilateral	Total
Ovarian sliding hernia	50 (36.2%)	35 (25.3%)	8 (5.8%)	9 (67.3%)
Nuck canal cyst	11 (8%)	5 (3.6%)	1 (0.7%)	17 (12.3)
Intestine	7 (5%)	3 (2.2%)	2 (1.5%)	12 (8.7%)
Lymphadenopathy	1 (0.7%)	0	0	1 (0.7%)
Hernia reduced under general anesthesia	8 (5.8%)	5 (3.6%)	2 (1.5)	15 (10.9%)
Total	77 (55.8)	48 (34.8%)	13 (9.4%)	138 (100%)

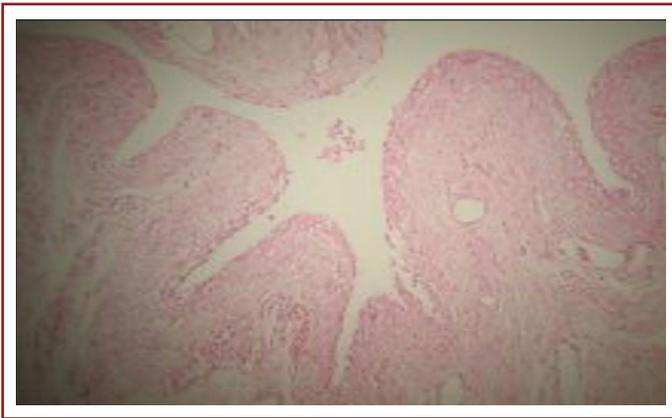


Figure 3. Nuck cyst wall with partially lined by a stratified squamous, transitional and columnar epithelium. In focal areas, moderately chronic active inflammation (HE, $\times 10$ magnification).

Discussion

Indirect inguinal hernia has an incidence of 0.8–4.4% in children and is 3–10 times more common in boys than girls. The first sign of inguinal hernia is incarceration in 10–20% of cases. The risk of incarceration is 12% for boys and 17.2% for girls. In this study, 15.9% could not be reduced. There may be evidence of the colon, small intestine, appendix, or Meckel diverticulum in the hernia sac, as well as the ovaries/Fallopian tubes and uterus in girls. The differential diagnosis should also include inguinal lymphadenomegaly, tumor, lipoma, cystic formations due to swelling, and a mass-occupying lesion in the groin^[3,5].

There is not enough information in the classic textbooks about the differential diagnosis, incidence, and treatment of CCN. CCN was first described by Dutch anatomist Anton Nuck Van Leiden in the 17th century. It is seen clinically in the inguinolabial region as a painless, mobile, non-abdominal mass. These clinical manifestations resemble those of an ovarian sliding hernia that cannot be reduced and CCN is often diagnosed as ovarian sliding hernia. CCN is mostly presented in the form of case reports in the literature^[6,7]. Among large series of female inguinal procedures, it has been reported an incidence of 0.76–1.03%^[8,9]. In this study, we detected CCN in 17 cases, which represented 2% (17/868) of all hernias and 12.3% (17/138) of hernias that could not be reduced. We think that the overall instance of CCN may not be sufficiently recorded or reported.

The diagnosis of inguinal hernia is usually made based on the clinical findings and a physical examination. The US is the most frequently used diagnostic tool for a complete differential diagnosis. Advantages include the ability to preliminarily define the hernia content and the relationship of the hernia to the abdomen, as well as observation of the

circulation, and the absence of radiation exposure^[10]. In this study of patients with an initial diagnosis of inguinal hernia that could not be reduced, only 34 patients underwent a preoperative US examination because it was either not possible or the surgeon did not request the US. The presence of CCN in two patients reported as ovarian sliding hernia/ovarian cyst suggests that CCN may not be distinguished in the differential diagnosis of inguinal hernia.

The treatment of inguinal hernia is surgical repair. Due to the high risk of incarceration in infants and children, most surgeons prefer to operate on pediatric hernias without delay^[11]. Surgery can be postponed for 24–48 h for a safer repair if the incarcerated inguinal hernia can be reduced. In hernias that cannot be reduced within 2 h, urgent surgery is recommended due to the risk of circulation impairment. Premature and low birth weight infants are typically operated on when they reach 2 kg just before they will be discharged from the neonatal unit due to the high risk of incarceration, while other infants <4 months of age are operated on within 2–3 weeks after the establishment of the diagnosis. Incarceration is most common in the first 6 months of life and is relatively rare after 5 years of age^[3].

The ovaries are commonly be found in the irreducible hernia sac of girls. There is no consensus among pediatric surgeons regarding the timing of surgical treatment of a sliding hernia of the ovary. There are surgeons who perform emergency surgery due to the possibility of incarceration and torsion of a herniated ovary, and there are surgeons who think that this possibility is very low and perform surgery under elective conditions^[12]. Imaging methods such as US, MRI, and CT can assist with confirmation of the diagnosis.

Studies in the literature generally include the pediatric age group and offer no precise recommendations regarding the preferred timing of surgical treatment of CCN^[8,9]. No complications have been reported in patients operated on at a more advanced age^[13,14]. In our study, no complications were observed in any of our CCN patients.

A hydrocelectomy is generally delayed until the age of 2 years in male patients, since the hydrocele may spontaneously regress^[15]. We believe that CCN, which is considered to be the embryological equivalent of a hydrocele, should be included in the differential diagnosis of female patients, and that there may not always be a need for urgent surgical treatment due to the possibility of spontaneous regression and the absence of organs or tissues in which circulation will be disrupted. Nonetheless, meta-analyses are needed to determine the ideal age for surgery.

Conclusion

Sliding hernia of the ovary was the most frequent finding (67.3%) in girls with the initial diagnosis of irreducible inguinal hernia, followed by CCN (12.3%). CCN mimics an inguinal hernia that cannot be reduced and therefore may be overlooked. CCN should be included in the differential diagnosis and advanced diagnostic methods should be used, given the significantly higher incidence compared with incarcerated inguinal hernia. The older age of CCN patients should be noted.

Ethics Committee Approval: On receipt of approval from the local Ethics Committee (11337-05.31.2019), a retrospective search of hospital information systems was performed to collect the data of female patients referred to surgery with the initial diagnosis of inguinal hernia in the pediatric surgery clinics of the Health Sciences University Umraniye Training and Research Hospital, İnönü University Faculty of Medicine Hospital, and Sivas Numune Hospital.

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