Cyst of the Canal of Nuck in adult females: A case report and systematic review

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Abstract. Defects in the Canal of Nuck are rare abnormalities of the female genitalia, which are typically detected and repaired in young age. In the present report, a case of a Nuck cyst in a 40-year old female patient is described. Additionally, the current literature concerning cases of women with hydrocele of Nuck canal was systematically reviewed. A total of 16 case reports of 16 patients with Nuck hydrocele (mean age of 35.18 years), have been reported to date. A right inguinal mass was noted in 13 patients (81.3%) whereas in 3 patients a left-sided mass was noted. The surgical approach was open in 13 cases and laparoscopic in 3 cases. Two cases underwent hydrocelectomy and inguinal ring ligation, whereas in 7 cases a simple cystectomy was performed. In 2 cases the round ligament was excised along with the hydrocele. In one of these 2, ligamentum rotundum necrosis and presence of a haemorrhagic cyst of the canal of Nuck were identified. Hernia repair and hydrocelectomy was performed in 5 cases. A cyst of the Canal of Nuck is relatively rare, but should be considered during the diagnosis of inguinal masses in female patients.

Introduction

Defects in the canal of Nuck are rare abnormalities of the female genitalia that are usually detected and repaired in young girls more frequently during the first 5 years of life (1). The first noted case defect in the Canal of Nuck dates back to 1691 by Anton Nuck (2). The failure of the canal to close after birth or within the first year of life in female infants can lead to formation of hydrocele or herniation of intraabdominal structures through the patent Canal of Nuck (1,3). Hydrocele of the Nuck canal, cyst of the Canal of Nuck or female hydrocele are equivalent terms for a rare developmental disorder of the reproductive system of women and accounts for a limited number of cases of benign painless or painful swelling in the inguinal region or even to the labia majora (4,5). Hydrocele of the Canal of Nuck constitutes a particular type of primary idiopathic hydrocele, the enlargement of which has been attributed to a defect of the secretory membranes resulting in an imbalance in secretion and absorption of fluids of the processus vaginalis (6).

Several reports of cysts of the Canal of Nuck are presented in literature and describe the symptomatology and surgical approaches used in their treatment (7-10). Although a rare pathology of the inguinal canal in female patients, it should be considered when diagnosing inguinal tumours in female patients. In the present report, a case of cyst of Nuck in a female patient, and a review of the current literature with special consideration on clinical presentation, diagnostic approach and management are presented.

Case presentation

A 40-year old female presented to our department with a painful mass in her right inguinal region. The swelling was first noticed two months ago, and the patient reported she never suffered from regional pain before. There was no history of local trauma, symptoms of nausea, vomiting or abdominal discomfort. Her body mass index was 27 kg/m^2. Her medical history was negative for any pathology and surgical procedures. She had two uncomplicated vaginal deliveries. At presentation, physical examination revealed a small palpable mobile lump in the right groin without overlying skin erythema or tenderness. Valsalva maneuver did not make the mass more prominent. There was an absence of incarceration or strangulation. Her abdomen was soft, non-distended and non-tender with no signs of bowel occlusion. Laboratory tests showed measured parameters were within the normal range. Magnetic resonance imaging (MRI) revealed a 3.5 cm well-defined, thin-walled cystic structure in her right groin (Fig. 1). There was no evidence of bowel loops, omentum or other solid structures

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within the mass. The diagnosis of a Nuck cyst was considered and the patient was operated on. The cyst was dissected from the round ligament and was completely excised. The defect of the internal inguinal ring was primarily repaired without the use of a mesh. Histology confirmed the presence of Nuck cyst. The patient's postoperative course was uneventful, and she was discharged the next day. One year postoperatively, the patient remained asymptomatic without any recurrence. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

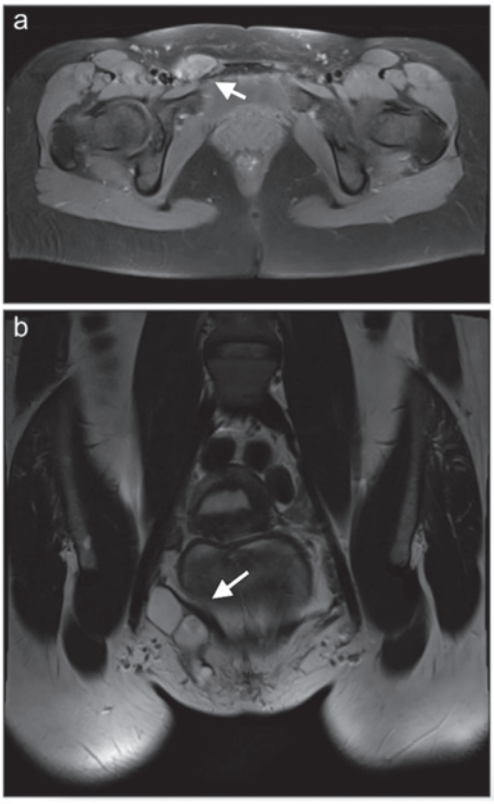

Figure 1. Magnetic resonance imaging showing a thin-walled cystic structure located in the right groin. (a) Axial view. (b) Coronal view.

Review of the literature

A systematic search of the literature for articles published between January 2000 and May 2019 was performed using the Medline (1966-2019), Scopus (2004-2019) and Google Scholar (2004-2019) databases along with the references in any articles. For all articles, the full-text was retrieved. Studies regarding adult women who were diagnosed with Nuck hydrocele were considered eligible for inclusion. A total of five studies were excluded from the present review (11-15). Pandey *et al* (11) and Safak *et al* (12) presented radiological outcomes of a case with Nuck hydrocele and thus excluded due to insufficient data. Noguchi *et al* (13) and Amu *et al* (14) were not considered eligible due the fact that the cystic structure in the Nuck canal was histologically confirmed as ectopic pregnancy and dermoid cyst, respectively, and not as hydrocele. Finally, Sala *et al* (15) included a 17-year old patient with Nuck cyst and was excluded due to age restrictions of the present study.

A total of 16 case reports of 16 patients with hydrocele of the Canal of Nuck, were included in the present systematic review (3-5,7-10,16-24); whereas 5 studies were excluded. The primary characteristics of the included studies including the primary symptoms on presentation, outcomes of clinical examination and imaging, the size of the mass and the type of procedure performed are presented in Table I. The mean age of patients included was 35.18±3.27 years and 13 (81.3%) of the patients were of reproductive age (18-45 years). Two of the included women were nulliparous; one of them presented with infertility and underwent simultaneous surgery for an enlarged ovarian cyst. Three women had one or more children. For the remaining 11 patients, data concerning parity was not available. A right inguinal mass was noted in the majority of the patients (n=13, 81.3%) whereas in the remaining thee patients a left-sided mass was noted. The enrolled patients were admitted with a groin swelling which was either painful in 5/16 cases or painless in the remaining 11/16 cases. In all except one case, preoperative imaging was performed; In six patients an ultrasound (US) was performed, preoperative computed tomography (CT) was performed in three cases, one patient underwent MRI, in one case a CT along with an MRI was performed, in four cases US and MRI were performed, and in one case US in combination with CT was performed. In seven cases, imaging suggested the presence of a cystic structure suggestive of the presence of hydrocele of the Canal of Nuck. Clinical examination revealed a reducible lump in the affected inguinal region in six women, and in eight cases, the mass was irreducible. In two cases, the nature of the mass was not specified.

All the included patients underwent surgery to examine the lesion and repair the defect. In 13 patients the surgical approach was open, and the other three patients underwent laparoscopy. Two cases underwent excision of the hydrocele and inguinal ring ligation, and in six cases a simple cystectomy was performed, which was followed by a vulva correction in one case. In 2 cases the round ligament was excised, with the hydrocele in one of them due to necrosis and presence of a haemorrhagic cyst. A hematoma within a Nuck hydrocele was found in one case and evacuated along with the hydrocelectomy. Hernia repair along with hydrocelectomy was performed in five cases (three laparoscopic and two open); among which, in two cases, a herniorrhaphy (no mesh repair) was performed, whereas three patients underwent a hernia repair with the use of a mesh. An additional inguinal ring ligation was performed in two patients; one laparoscopic and one open. Postoperative recovery was uneventful, and no postoperative deaths or complications were observed. According to 7 of the included studies, no recurrence of the lesion was reported in the follow up period, which ranged from 2-24 months after surgery (3,8,9,16,18,20,22). Data concerning hospital stay was reported in 4 studies with a range of 20-72 h (7,10,18,20).

Discussion

The Canal of Nuck runs through the inguinal canal adjacent to the round ligament and is considered the female analogue of the processus vaginalis in males (25). Normally, the Canal of Nuck is obliterated within the first year of life. Failure of the Canal to close during that period in female infants can result in Nuck hydrocele or herniation of intraabdominal structures through the patent Canal of Nuck (1). Thus, failure of closure is typically

Table I. Published studies included in the literature review.

Author, year (Ref)	Primary symptom; physical examination	Imaging modality	Size, cm	Surgical approach, Lap/Open	Type of repair
Bhattacharjee and Ghosh, 2006 (3)	Left painless swelling; reducible, soft, cystic, non-tender	U/S: Encysted echofree lesion in the left inguinal canal.	7.5x5.0	Open	Hydrocelectomy and wound closure
Caviezel *et al*, 2009 (4)	Right painless swelling; irreducible, increased volume in standing position reduced by manual compression	U/S and MRI: Anechogenic cystic mass with. thin wall	5.0x5.0	Open	Hydrocelectomy and round ligament excision
Jagdale *et al*, 2012 (16)	Right painful swelling; irreducible tender, cystic and fluctuant	U/S: Well-defined, avascular, oval, anechoic cystic swelling within the inguinal canal.	4.3x2.6	Open	Hydrocelectomy and NMR
Ferreira *et al*, 2017 (17)	Painless vulval swelling; irreducible soft fluctuant sausage-shaped mass	U/S: Well-defined hypoechoic elongated mass with 5.5 cm of long axis, septated, extending from the superficial inguinal canal to labia majora.	4.0	Open	Hydrocelectomy and vulva correction
Karapolat *et al*, 2018 (18)	Right painless swelling; irreducible mass of medium hardness with smooth surface and limited mobilization	U/S: Thick-walled cystic lesion with thin internal septae. MRI: cystic lesion with lobulated contours T1A hypointense and T2A hyperintense intensities neighboring.	2.5x5.0, 3.5x4.7	Open	Hydrocelectomy and inguinal ring ligation
Matsumoto *et al*, 2014 (7)	Left painless swelling; reducible	U/S: Hypoechoic and homogeneous without solid components. MRI, simple cystic lesion, which appeared to be in contact with the left ovary connected at its base with the parietal peritoneum.	4.5	Lap (TEP)	Hydrocelectomy and MR
Ozel *et al*, 2009 (19)	Right painless swelling for 3 months; irreducible fluctuant sausage shaped	U/S: Tubular cystic structure with thin internal septae not change its shape when compressed by the transducer with no abnormal vascularity. MRI: Well defined lobulated tubular mass which was hypoin tense on T1-weighted images and hyperintense on T2-weighted image	6.0x4.0x1.5	Open	Hydrocelectomy
Patnam *et al*, 2016 (9)	Painless right swelling for 3 months; irreducible positive fluctuation. No expansible cough impulse, no peristaltic activity and no abnormal vascularity associated with the swelling	U/S: Elongated, anechoic fluid collection, not change in size on Valsalva maneuver, nor did it suggest a communication to the peritoneal cavity. CT: Well-defined, peripherally enhancing thin walled tubular cystic structure extending from the right iliac fossa along the course of the round liga ment through the right inguinal canal to the ipsilateral labia	6.3x3.4x2.0	Open	Hydrocelectomy and wound closure

Table I. Continued.

Author, year (Ref)	Primary symptom; physical examination	Imaging modality	Size, cm	Surgical approach, Lap/Open	Type of repair
Qureshi and Lakshman, 2014 (8)	Left painful swelling for 1 month; irreducible, oval, tender, cystic and fluctuant swelling	U/S: Left inguinal hernia, with well-defined, oval, anechoic cystic swelling within the inguinal canal	4.0x3.0	Lap	Hydrocelectomy and MR
Yen *et al,* 2001 (20)	Right painless mass; reducible above labia major, reduced in supine position	Not performed	N/A	Lap (IP)	Hydrocelectomy, inguinal ring ligation and NMR
Uzun *et al,* 2017 (10)	Right painful swelling for 2 days, 4 days following vaginal delivery	CT: Inguinal hernia, with a suspicion of herniation of the adnexal organs	N/A	Open	Hemorrhagic cyst and necrotic liga mentum rotundum excision
Zawaideh *et al,* 2018 (5)	Right painful swelling.	U/S: Oval anechoic mass	N/A	Open	Hydrocelectomy
Kim *et al,* 2016 (21)	Right painless swelling for 4 months; reducible	CT: Cystic mass in the inguinal canal. MRI: Cystic mass in the inguinal canal included thin septa	7.1x3.8	Open	Hydrocelectomy and high inguinal ring ligation
Sethi and Patel, 2016 (23)	Right painless swelling for 1 year; reducible fluctuant, and minimally tender mass, without a bruit or thrill	CT: Oval fluid collection in the right inguinal region extending to the right labia majora.	11.7x4.9x3.6	Open	Hydrocelectomy
Kono *et al,* 2015 (22)	Right painless swelling for 2 years; palpable mass	MRI: Irregularly shaped cystic mass lesion and a smaller cystic lesion and fluid collection evident in the right side of the pelvic cavity.	4.8x3.7	Open	Hydrocelectomy (outside lesion), inguinal ring ligation, MR
Ryan *et al,* 2009 (24)	Right painful swelling for 2 days; irreducible, ecchymosis, tender, firm, palpated superolateral to the pubic tubercle	U/S: Mass within the right groin, with both cystic and solid components present with the suggestion of the possibility of bowel contents.	N/A	Open	Hematoma evacuation and hydrocelectomy

N/A, not available; CT, computer tomography; U/S, ultrasound; MRI, magnetic resonance imaging; Lap, laparoscopic; TEP, total extraperitoneal; IP, intraperitoneal.

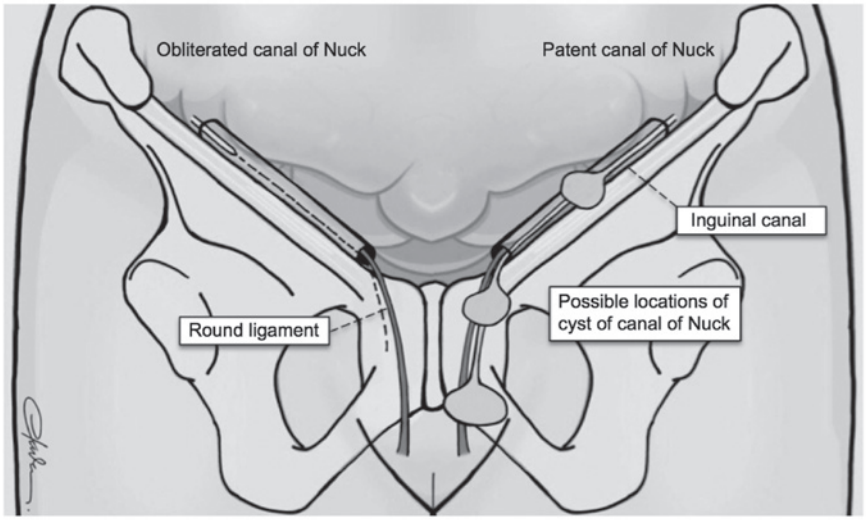

Figure 2. Anatomy of the inguinal canal with a physiologically obliterated Canal of Nuck and the potential sites of the cyst through the canal when it remains patent.

detected in childhood. Due to its rarity, accurate information regarding the exact prevalence of hydrocele during childhood is not available. Akkoyun *et al* (26) reported that only 0.76% of girls <12 years exhibited hydrocele of Nuck among their study population. A comparable prevalence of Nuck hydrocele (0.74%) was also reported by Papparella *et al* (27), who reviewed 353 female patients, aged 1-14 years, with inguinal swelling. Literature regarding Nuck hydrocele in adulthood are even more scarce. A systematic review performed in the present study revealed a total of 16 cases of adult patients who underwent surgery for Nuck cyst.

Fig. 2 schematically presents the anatomy of the inguinal canal with a physiologically obliterated Canal of Nuck and the potential sites where the cyst may be recognized when the canal is patent. At clinical examination, the cyst of the Canal of Nuck is frequently described as a painless or mildly painful irreducible or reducible mass in the inguinal region, which typically extends to the labia majora, and does not expand when performing the Valsalva manoeuvre (23). In approximately one-third of patients an associated inguinal hernia is present. Consistent with this, nearly one-third of the patients included in the present study underwent a simultaneous hernia repair. Differential diagnosis includes inguinal hernia, enlarged lymph nodes and soft tissue tumours such as lipomas, leiomyomas and endometriosis of the round ligament (28).

A cyst of the Canal of Nuck is frequently misdiagnosed as inguinal hernia in females and is only correctly diagnosed intraoperatively. Therefore preoperative imaging is crucial for facilitating diagnosis and further guiding therapeutic options. All except one of the included cases included in the systematic review underwent preoperative imaging with US, CT, MRI, or a combination of these. High-resolution real-time sonography serves as an inexpensive and accurate modality for differentiating hydrocele of the Canal of Nuck from the aforementioned conditions. In an ultrasound scan, Nuck cyst appears as a thin walled, tubular or dumbbell shaped, well defined, anechoic or hypoechoic, unilocular or multilocular cystic structure (19,22). In a colour Doppler scan, the Nuck cyst does not show any internal vascularity (28,29). Additionally, an MRI may also be performed, particularly in complicated cases, such as those involving additional pathologies where hydrocele usually appears as a well-defined, thin-walled cystic lesion in hypointense on T1-weighted and hyperintense on T2-weighted series (1). Imaging with MRI allows for good visualization of the anatomic structures surrounding the cyst, communication between the cyst and the peritoneal cavity and the extension of the cyst of the Canal of Nuck (22). However, despite the utility of imaging in differential diagnosis, surgery along with histological and immunohistological analysis of the excised mass is required for a more conclusive diagnosis of a Nuck cyst.

Another issue that should be addressed is the association of the pathology of the Canal of Nuck with fertility. In the present case report as well as in the published literature, there was only one association with infertility; a nulliparous woman of reproductive age who underwent simultaneous ovarian cyst excision and repair of patent Canal of Nuck (20). Nonetheless, infertility in this case was not attributed to the Nuck canal pathology. Postoperative courses of all published cases are uneventful. Unfortunately, data concerning postoperative fertility was not available for any of the reported cases. Other pathologies of the Canal of Nuck such as ovary herniation or endometriosis of the canal may result in infertility in young females (30).

Surgical management of a cyst of the Canal of Nuck includes open or laparoscopic excision of the cystic structure with concomitant closure of the inguinal internal defect primarily with the use of a mesh (24,31,32). The appropriate surgical approach is tailored based on the extent of the disease, the accuracy of preoperative diagnosis and the co-existence of an inguinal hernia. In the case of concomitant identification of an inguinal hernia, an additional hernia repair with or without mesh placement can be safely performed. Furthermore, as described by Ferreira *et al* (17), an additional vulva correction may be indicated in cases of mass extension to the labia majora.

To the best of our knowledge this is the first literature review which presents a cumulative report of cases of adult females with Nuck cyst. The risk of potential loss of relevant literature was eliminated by performing a thorough search of the current literature. Due to limited data from case reports and

small case series, the actual prevalence of Nuck cyst could not be precisely estimated. The significant heterogeneity among the included studies along with the lack of mention of certain parameters by some authors were additional limitations. In conclusion, the cyst of the canal of Nuck is a rare condition, but it should be included in the differential diagnosis list of inguinal tumours in female patients.

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Availability of data and materials

The datasets used and/or analysed during the present study are available from the corresponding author on reasonable request.

Authors' contributions

APr and NM conceived and designed the study. APr, APa, DS, CN and ES acquired, analysed and interpreted the data. APr, APa, DS, CN, NM and ES drafted and revised the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

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