

Adnexal Sliding Hernia in an Infant: Clinical-Anatomical Case Report

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Abstract

We present a case report of laparoscopic herniorrhaphy in an infant with ovarian prolapse. Using this case study, we demonstrate the role of the ovarian ligamentous apparatus. A structure appearing like the male gubernaculum was identified. The anatomical and functional role of this "gubernaculum" will be the subject of discussion. Further detailed laparoscopic examinations are indicated to better understand the ligamentous anatomy of ovarian prolapse.

Keywords

Inguinal Hernia, Ovarian Prolapse, Ovarian Descensus, Sliding Hernia, Gubernaculum

1. Introduction

The diagnosis and treatment of pediatric inguinal hernia is considered to be one of the core competencies of pediatric surgery. It is imperative that pediatricians and neonatologists possess a comprehensive understanding of the symptoms and potential complications associated with this condition. In the vast majority of cases observed in childhood, an indirect or lateral inguinal hernia is present, with a male preponderance that exceeds 80%. Furthermore, 3% - 5% of newborns and 13% of premature babies with a gestational age of less than 33 weeks are found to develop an inguinal hernia [1] [2]. In the presence of a pediatric inguinal hernia, the internal inguinal ring and inguinal canal act as a hernial orifice. In the terminology of congenital inguinal hernia in girls, the eponymous term Nuck's canal is used. The hernia sac corresponds to the persistent, open processus vaginalis peritonei. This normally obliterates towards the end of the fetal period or in the first

few months postnatally. The definitive surgical treatment consists of closing the inner inguinal ring or the hernia sac. Both open surgery and minimally invasive procedures are accepted and highly effective methods. The laparoscopic procedure offers some advantages, particularly in girls [3]-[5]. A prolapsed ovary is the most common presentation of inguinal hernia in female infants. Compared to open surgery, the laparoscopic herniorrhaphy allows a more differentiated view of the anatomy of the groin and internal genital tract.

It is controversial whether the ligamentous apparatus plays a crucial role as a guiding structure in fetal ovarian descent. Furthermore, there is an ongoing debate as to whether the male gubernaculum and the female round ligament are homologous structures. In principle, the gubernaculum can be defined as the ligamentous connection between the gonad and the deepest point of the descensus.

According to Dorland's Illustrated Medical Dictionary [6], the gubernaculum testis is defined as the "fetal ligament attached to the inferior end of the epididymis, at its other end, the bottom of the scrotum". It is present during and is thought to guide the descent of the testis into the scrotum. It is called Hunter's ligament after the eminent anatomist and surgeon who first described it. In this communication we describe the laparoscopic appearance of the ligamentous suspensory apparatus.

2. Case Report

This paper presents a case study of a former premature baby girl with ovarian prolapse. The baby was delivered by vias naturales 8 weeks before the due date, and was four months old at the time of presentation. The birth weight (z = 0.02) and the actual weight were both within the age- and gender-related normal range for a corrected age of two months. Upon first presentation, a bulging of the cranial portion at the left labium was immediately apparent. No further symptoms were present. The prolapse had obviously been there for some time. The infant was fed on breast milk and later on formula. A unilateral prolapsed ovary was confirmed by ultrasound. An almond-shaped resistance was palpaple subcutaneously. A Doppler examination revealed persistent blood flow in the contents of the hernia sac. An attempt was made at laparoscopic percutaneous extraperitoneal herniorrhaphy (LPEC) [7].

A 5 mm camera (Storz^{*}, Tuttlingen, Germany) was passed through the umbilical ring via an open approach, after which a low pressure 8 mmHg capnoperitoneum was established. A closed deep inguinal ring was seen on the right side, and the round ligament of the uterus was in contact with the deep inguinal ring. It was note-worthy that the infantile uterus was deviated to the left side by traction. The left deep inguinal ring and the vaginal process were found to be wide open. Two ligamentous structures and the fallopian tube entered the left deep ring (see Figure 1(a)). Outside the inguinal canal the ligamentum ovarium proprium (synonymously utero-ovarian ligament) was observed to come from the medial side and run parallel to the Fallopian tube. Additionally, the stretched cranial suspensory ligament of the ovary

originated from cranial and extended laterally along the edge of the pelvis into the hernia sac. The prolapsed ovary was reduced by means of a 3 mm grasper, which was inserted from the inside and manipulated manually from the outside. The well perfused but edematous ovary and the funnel of the ovarian tube became visible (**Figure 1(b)**). At the base of the ovary, a third ligament with a short appearance was identified, which was inserted at the lateral region of the inverted hernia sac. This ligament resembled the gubernaculum according to the topography and where it is located on the female gonad (**Figure 2(b)**). The deep internal ring was closed with a modified 2×0 purse-string suture (Ethibond[®], Ethikon, Norderstedt, Germany). One year following the surgical procedure, the mother reported no complications when contacted via telephone. Subsequently, however, the family was no longer contactable for further follow-up.



(a)



(b)

Figure 1. Laparoscopic view from above of the left deep inguinal ring. (a) Three structures herniate over the posterior edge of the deep inguinal ring (from left to right): (1) the cranial suspensory ligament of the ovary (LSO), (2) the round ligament of the uterus (ligamentum ovarii proprium, LO) and (3) the uterine tube arising from the uterine horn (UH, uterine horn). Arrow pointing in the direction of the entrance to the inguinal canal. (b) The ovary (O) is pulled back into the pelvic cavity. Now the third ligamentous structure comes into view: the "gubernaculum" (star). It runs from underneath the ovary through the deep inguinal ring (IR) and attaches within the sac. The tip of the arrow at the base is directed towards the tiny fimbriae of the uterine tube.



Figure 2. Schematic drawing of the Prolapse of the ovary and the Fallopian tube through the inguinal canal. (a) The uterine tube (blue) is accompanied by the LSO (green), (2) LO (red). The short "gubernaculum" situated below the ovary are not depicted from above (**Figure 2(a)**). It appears to originate separately from the ligamentum teres uteri. (b) The distal part of the round ligament of the ovary, otherwise known as the "female gubernaculum", was rendered visible during the process of pulling reduction.

3. Discussion

A prolapsed ovary is the most common presentation of inguinal hernia in female infants. The only identifiable predisposing factor for the development of inguinal hernia was prematurity. In asymptomatic cases with ovarian prolapse, herniotomy should be performed during the next days. Pain, vomiting and local edema or redness are signs of ischemia and require emergency surgery within hours. However, it has to be noted, that Strangulation of the contents of the hernia sac or torsion are the most common emergencies. The presented case report focusses on ovarian descensus, developmental mechanics and in particular on the complete ligament anatomy. A discussion of both the practical and epistemological aspects is required. From a biomechanical perspective, the ligamentum teres uteri can be incorporated into the inner purse string suture for laparoscopic hernia repair. This maneuver will enhance the integrity and stability of the inguinal ring closure. Unfortunately, the terminology and anatomy of the ovarian ligaments during descensus is inconsistent and sometimes confusing. In relation to the ovary, the current anatomical nomenclature describes two ligaments, the ligament of the ovary (LO) or synonymously the utero-ovarian ligament, and the suspensory (LSO). The round ligament of the uterus (lig. teres uteri) also plays an important role [8].

A schematic representation of the topography of the tubular structures that herniate into the deep inguinal ring is presented in Figure 2(a) and Figure 2(b). The term "gubernaculum" employed in the illustrations depicting the ligament between the ovary and the hernia sac is to be comprehended as a working term. The rationale underlying this designation is elucidated in the ensuing discourse.

The classic textbook by Patten (1953) describes the diaphragmatic ligament of the mesonephros (corresponding to the later suspensory ligament) and, caudal to the ovary, the "inguinal ligament", which merges into the round ligament of the ovary. These ligaments form the guardrail of the ovarian descensus. In addition, it was traditionally thought that the distal part of this round ligament of the ovary (genitoinguinal ligament) was anchored in the connective tissue of the labium majus [9]. However, Hutson and coworkers (2016) found that the round ligament does not extend to the labium majus [10]. Anatomical studies in adults describe three different types of distal insertions of the round ligament [11]. Our observation confirmed an insertion at the internal surface of the hernia sac. In addition, previous research on the origin of ovarian hernia suggested that the processus vaginalis is formed nearby the "gubernaculum". Stretching of the open processus vaginalis was thought to be responsible for displacing the end of the round ligament to the side of the hernia sac—Hutson's sliding hernia theory. It should also be considered that non-androgenic hormone controlled "overdescensus" may contribute to the development of ovarian hernia [12].

On the basis of the study presented, it cannot be whether "overdescensus" played a causal role. As a result of our observation we can state that the prolapse of the adnexa is associated with two ligaments and the Fallopian tube entering the deep inguinal ring. The short distal part of the round ligament of the ovary "gubernaculum" is visible after reduction. Shortening of this "gubernaculum", as described by Kuyama *et al.*, 2019, may contribute to the displacement of the ovary. In the case of an open process, neither the suspensory ligament nor the utero-ovarian ligament is sufficient to hold the ovary in its orthotopic position [13].

Finally, it should be noted that the methods of laparoscopic repair of pediatric inguinal hernias are constantly improving. Compared to the LPEC method used in this case, a procedure with additional laparoscopic hernia sac transection has proven to be particularly effective [14]. It is no longer considered that the open inguinal herniotomy is the sole gold standard in the treatment of girls. However, open access via a small gross incision is still a highly accepted method due to the following reasons: it is a rapid procedure, it is simple, it requires few resources

¹This ligament is not identical with the inguinal ligament according to the recent nomenclature.

and ipsilateral relapses are very rare [15] [16].

4. Conclusions

Our single case observation shows that formally a morphological analogue reminiscent of the male gubernaculum. This did not simply appear as a continuation of the ligamentum suspensorium ovary or the ligamentum teres uteri (round ligament). Rather, an independent ligament was found between the ovary and the lowest point of the hernial sac. Functionally, however, a homology to the male descensus testis is unlikely. In this specific context, the use of the term "gubernaculum" for girls does not get to the core.

Further on, more laparoscopically based case series are suitable for clarifying the anatomy, which may also differ depending on the case. However, investigations into the ligament anatomy also seem to make sense in the open procedure.

Terminology [6] [8] [17] and Abbreviations

Adnexa uteri: Uterine appendages (e.g. uterine tube, ovary)

Uterine tube and uterine horn: Fallopian tube. The proximal anatomical opening of the uterine tube is the uterine horn (UH) of the uterus. The uterine tube is named after the Italian renaissance anatomist and surgeon Gabriele Fallopio (Modena, Italy).

Deep inguinal ring, internal inguinal ring (IR): Internal entrance to the inguinal canal. This is where the inguinal canal is closed during the laparoscopic procedure.

Canalis inguinalis: The passage through the aponeuroses and the muscle of the abdominal wall, superior to the inguinal ligament. This canal extends from the deep inguinal ring to the superficial inguinal ring, facilitating the transmission of the round ligament in the female.

Patent processus vaginalis peritonei: Canal of Nuck. The eponym refers to the open vaginal process in females. The processus vaginalis peritonei in the female is usually obliterated after birth, but may persist. Anton Nuck was a professor of anatomy and medicine in Leyden, Holland.

Gubernaculum testis: Hunter's gubernaculum. It is a structure that plays a pivotal role in the guidance of the descensus testis. This structure is attached to the inferior aspect of the epididymis, and at its other extremity, it connects to the bottom of the scrotum. It is hypothesized that the gubernaculum assists in the descent of the testis into the scrotum. The gubernaculum typically atrophies over time. The ligament is named after the famous Scottish-English anatomist and surgeon John Hunter (Long Calderwood and London).

Ligamentum suspensorium ovarii (LSO), suspensory ligament of the ovary: It is a structure that can be found in the peritoneal cavity. It is located laterally and superior to the ovary, and is connected to the broad ligament. This structure contains ovarian vessels and nerves, and it passes upwards over the iliac vessels.

Ligamentum ovarii proprium (LO), ovarian ligament: The structure under

consideration is a musculofibrous cord located in the mesovarium portion of the broad ligament. It connects the ovary to the upper part of the lateral margin of the uterus, situated just below the attachment of the uterine tube. The structure is also known by the synonyms utero-ovarian-ligament or ligamentum uteroovaricum.

Ligamentum teres uteri, round ligament of uterus: The structure is attached to the uterus at the point where the proper ovary ligament attaches. It traverses the anterior layer of the broad ligament and exits through the inguinal canal, where it becomes attached to the inner aspect of the skin of the labium majus. However, it should be noted that this proposal has been the subject of recent revision by researchers in the field.

Authors' Contributions

Conceptualisation: RBT. Writing of the original draft: RBT. Writing of final manuscript: RBT, GC, MN.

Data Availability

All data generated or analyzed for this case report are included in this published article.

Ethical Approval and Concept of Participation

Due to the retrospective nature of the case report and the lack of patient identifiability, ethical approval is not required.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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