doi: 10.48095/cccg2024411

Interlabial masses in newborn girls

Interlabiální útvary u novorozených dívek

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Summary: An interlabial mass in newborn girls is diagnosed usually after birth or during the first days or weeks of life. According to various studies, its prevalence ranges between 1 : 500 and 1 : 7,000 newborn girls. A mass in the vaginal introitus or between the labia majora can cause a diagnostic dilemma and may be suspected even of ambiguous genitalia. Interlabial masses of different etiologies present clinically similar, and therefore, can be misdiagnosed. The most common causes of an interlabial mass in a newborn are hymenal and paraurethral cysts, both of which present as thin-walled spherical formations filled with golden fluid. When diagnosing a cystic interlabial tumor, it is necessary to particularly exclude a non-perforated hymen with hydrocolpos and prolapse of an ectopic ureterocele. In the differential diagnosis, prolapse of the urethra, rhabdomyosarcoma of the vagina or cervix, urethral or vaginal polyps, and extremely rare conditions such as genital prolapse or duplicate rectum cannot be omitted. A newborn girl with an interlabial formation should be examined by a pediatrician, gynecologist, surgeon, or urologist depending on the nature of the clinical findings. Once the etiology of an interlabial mass is identified, expectant management or surgery should be chosen. Early surgical treatment of hydrocolpos and prolapse of a ureterocele can prevent lower urinary tract obstruction and life-threatening renal damage.

Key words: interlabial mass – hydrocolpos – hymenal cyst – newborn – paraurethral cyst – prolapsed ectopic ureterocele – rhabdomyosarcoma – urethral prolapse – genital prolapse

Souhrn: Interlabiální tumor je u novorozených dívek diagnostikován v prvních dnech, resp. týdnech života. Vyskytuje se s prevalencí 1 : 500 až 1 : 7 000 novorozených dívek. Tumor ve vaginálním introitu, resp. mezi velkými stydkými pysky může způsobit diagnostické obtíže nebo vytvářet dojem genitálu nejasného pohlaví. Interlabiální útvary různé etiologie jsou si svým vzhledem podobné, což může vést k diferenciálně-diagnostickým omylům. Nejčastější příčinou interlabiálního tumoru jsou u novorozených dívek hymenální a parauretrální cysty, které se projevují jako tenkostěnné kulovité útvary vyplněné zlatavě zbarvenou tekutinou. V případě cystického interlabiálního útvaru je nutné vyloučit zejména atrezii hymenu s hydrokolpos a prolaps ektopické ureterokély. V diferenciální diagnostice je nutné pomyslet na prolaps uretry, rhabdomyosarkom pochvy a děložního hrdla, uretrální či vaginální polyp a u novorozence extrémně vzácný prolaps dělohy a poševních stěn nebo duplikaturu rekta. Novorozené dívky s interlabiálně lokalizovaným tumorem by měly být podle klinického nálezu vyšetřeny pediatrem, gynekologem, chirurgem nebo urologem. V závislosti na etiologii interlabiálního útvaru je možné zvolit expektační postup nebo chirurgickou léčbu. Včasná operace může zejména v případě hydrokolpos a prolapsu ektopické ureterokély zabránit obstrukci dolních močových cest a z toho plynoucímu zdraví ohrožujícímu poškození ledvin.

Klíčová slova: interlabiální tumor – hydrokolpos – hymenální cysta – novorozenec – parauretrální cysta – prolaps ektopické ureterokély – rhabdomyosarkom – prolaps uretry – prolaps dělohy

Introduction

Examination of the external genitalia is an integral part of the postnatal newborn's examination and later pediatric checks. An interlabial mass is detected either immediately after birth or by parents or a pediatrician in the first days or weeks of life. The prevalence of an interlabial mass in newborn girls varies between 1 : 500 and 1 : 7,000 [1,2]. A mass presenting in the vaginal introitus between the labia majora can cause a diagnostic dilemma and may be suspected of ambiguous genitalia. A pathological interlabial formation concerns both parents and pediatricians, and therefore, early postnatal gynecological examination following initial pediatrician examination should be performed to dispel diagnostic uncertainty.

The most common causes of an interlabial mass in a neonate are hymenal and paraurethral cysts, both of which present clinically as thin-walled spherical formations filled with golden fluid [2,3]. In the differential diagnosis, prolapse of the urethra, rhabdomyosarcoma of the vagina or cervix, urethral or vaginal polyps, and rare conditions such as genital prolapse or duplicate rectum cannot be

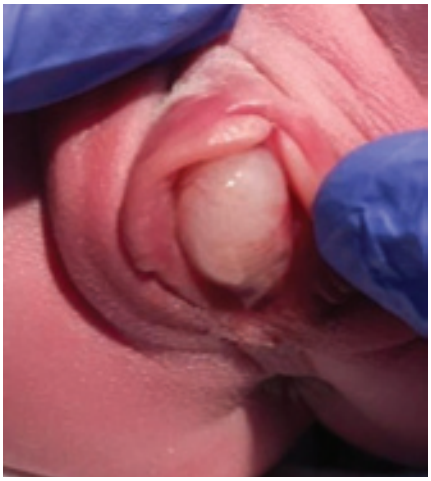

Fig. 1. Hymenal cyst in 1-day-old newborn.
Obr. 1. Hymenální cysta u 1denního novorozence.

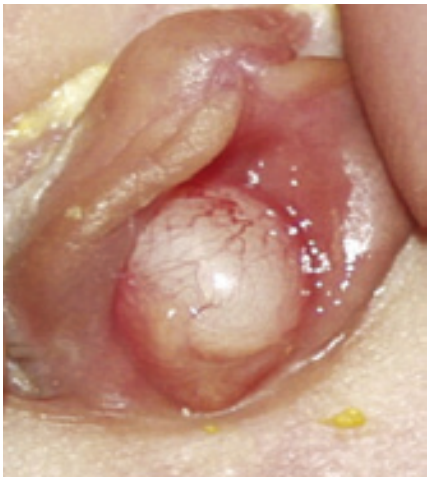

Fig. 2. Imperforate hymen with hydrocolpos.
Obr. 2. Neperforovaná panenská blána s hydrokolposem.

omitted [4]. All of the above causes of interlabial formations are strikingly similar and can be misdiagnosed.

In this article, the causes of interlabial masses in newborn girls are described, including their individual characteristics, typical clinical signs, optimal examination methods, and appropriate treatment approaches.

Hymenal cyst

Hymenal cyst is a thin-walled spherical formation between the labia majora adjacent to the hymenal edge containing a golden-colored fluid. It may rarely cause lower urinary tract obstruction. Within a few weeks after delivery, it usually disappears spontaneously without intervention [3]. In a majority of cases, simple physical examination is enough to differentiate a hymenal cyst from other types of interlabial masses requiring treatment. In a girl with a hymenal cyst, normal position of the external urethral meatus and vaginal introitus are confirmed. The benign nature and likelihood of spontaneous resolution are the reasons why surgical treatment of a hymenal cyst is not necessary [5].

A hymenal cyst in 1-day-old newborn girl is shown in Fig. 1. The cyst was connected by a peduncle to the upper right medial margin of the hymen. On day 3 of life, it spontaneously ruptured and emptied itself without the need for surgery.

Paraurethral cyst

The incidence of paraurethral cysts varies between 1:500 and 1:7,000 in newborn girls [1,4,6]. These types of cysts develop from the periurethral glands of the distal urethra; however, the exact etiology remains unknown. They are located lateral to the urethra and displace the urethral meatus contralaterally [4,5]. Paraurethral cysts are usually asymptomatic, but sometimes may cause an abnormal urinary stream or urethral obstruction [3,7]. During the first month of life, a paraurethral cyst usually manifests itself as a small (0.5–2 cm in diameter) yellowish or whitish interlabial cystic formation with dilated capillary vessels on its surface. The diagnosis is based on physical examination. Ultrasound examination is the method of choice in urinary outflow obstruction with diagnostic uncertainty, particularly if the urethral or vaginal meatus is masked by a cystic mass [1].

Either expectant or surgical approaches should be used in the treatment of paraurethral cysts. As they are usually asymptomatic and spontaneous regression is expected, a waiting approach may be recommended [4]. The interval to spontaneous resolution is variable, and according to the experience of different authors, it can range between a few days and many months of life [1,4]. Surgery such as incision, needle aspiration, excision, and marsupialization may be performed to treat urethral obstruction or otherwise symptomatic paraurethral cysts [4,6–8]. Early drainage of a paraurethral cyst could be performed to reduce parental anxiety. The risk of recurrence after surgery is low [1].

Imperforate hymen with hydrocolpos

Imperforate hymen is the most common obstructive congenital malformation of the female genital tract with an incidence of 0.014% to 0.2%. A girl with an imperforate hymen is usually asymptomatic until puberty. Then, accumulated cervical and vaginal secretions, along with menstrual blood, may distend the vagina and uterine cavity causing cyclic pelvic pain with amenorrhea [9–11].

When stimulated by maternal estrogens, the uterine and vaginal mucosa of the fetus may produce secretions and create hydrocolpos. The estimated incidence of neonatal hydrocolpos is 0.006% [12]. The diagnosis of an imperforate hymen is based on physical examination (Fig. 2). A shiny hymenal membrane bulging from accumulated secretion and protruding from the vaginal introitus is a typical clinical sign [11]. In a newborn with hydrocolpos, abdominal distension with bulging of the lower abdomen may also be present [13]. The diagnosis of an imperforate hymen is confirmed by abdominal ultrasonography showing a fluid-filled distended vagina (hydrocolpos), in severe cases also uterine cavity (hydrometra) and displacement of the uterus, bladder, or rectum [14]. Magnetic resonance imaging should be performed to rule out other genitourinary malformations [15].

This increases the risk of a newborn's morbidity, but the prognosis of girls with

an isolated imperforate hymen is generally good [16]. Urinary retention is a rare but serious consequence of hydrocolpos due to compression of the lower urinary tract with signs of obstructive uropathy such as hydroureter and hydronephrosis [11]. Therefore, early detection of hydrocolpos may prevent life-threatening kidney damage [10,17].

Although an imperforate hymen is a rare cause of hydronephrosis, ultrasonography of the urinary tract should be performed in any neonate with this congenital anomaly to avoid complications due to the late detection of hydronephrosis [18]. Symptomatic girls should undergo cross-incision hymenotomy with evacuation of the hydrocolpos [11,14].

Prolapsed ectopic ureterocele

Ectopic ureterocele is a congenital anomaly with cystic dilatation of the intravesical or extravesical part of the ureter. Although the majority of ectopic ureteroceles remain in their orthotopic position, some of them may prolapse through the urethral lumen during micturition causing sudden, intermittent, or permanent retention of urine. The incidence of a ureterocele ranges from 1:5,000 to 1:12,000 in female-sex infants. However, a prolapse of an ectopic ureterocele affects less than 5% of them [19,20].

The surface of a ureterocele is covered by vesical mucosa. Therefore, the prolapsed tissue is pink-colored, if detected early. However, duration of the prolapse increases the risk of ischemia of the prolapsed tissue, and the color of the ureterocele changes to bright red and bluish-purple (Fig. 3). When a ureterocele prolapses through the urethra, the urethral meatus is invisible. If the urethra is obstructed only partially, urine can be seen to flow around the ureterocele. The urethral meatus is central and above the prolapsed ureterocele, while the vaginal introitus, which is usually overlapped by the mass of the ureterocele, is located caudally and below [5,21,22].

Because a prolapsed ureterocele may cause obstruction of the lower urinary tract, the primary aim of treatment is to ensure drainage of urine. If the prolapsed mass is only manually pushed back into the bladder, it may recur early. In such cases, interlabial incision or endoscopic puncture with the reposition of the prolapsed sac could be an optimal emergency treatment modality [21].

The majority of female sex cases have a duplex renal system. Therefore, if a prolapsed ectopic ureterocele is suspected, concurrent urinary tract anomalies should be excluded. In an extravesical type of ureterocele, vesicoureteric reflux should be examined. To select an appropriate method of treatment, the extent of a ureterocele should be assessed using imaging methods [19,20].

Vaginal rhabdomyosarcoma (sarcoma botryoides)

Rhabdomyosarcoma is the most common soft tissue sarcoma in pediatric and adolescent populations. About 18–22% of cases are located in the genitourinary tract [23,24]. The average age at the time of diagnosis is less than 2 years, but rhabdomyosarcoma is rarely detected immediately after the birth [5,25].

Sarcoma botryoides presents usually as a large grape-like cluster of a pearly grey mass protruding from the vagina. Vaginal bleeding, abdominal pain, urinary incontinence, frequent urination, and constipation may also occur. Visual inspection is usually sufficient to confirm diagnosis because its clinical appearance is very characteristic [5,23]. In a majority of cases, staging examinations and biopsy with a histopathological analysis should be performed. In a minority of women with a small well-circumscribed tumor, primary surgical resection with no functional and cosmetic consequences should be performed. Rhabdomyosarcoma is sensitive to chemotherapy and radiotherapy. Brachytherapy is considered a good (and effective) alternative to external beam radiation because of decreased morbidity. A 10-year survival is achieved in 90% of patients. However, survival depends on the age of children at the time of detection because in those younger than 1 year, 10-year survival is lower [26].

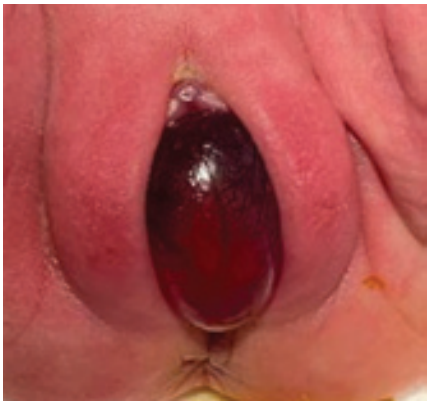

Fig. 3. Prolapse of an ectopic ureterocele in a10-day-old girl.
Obr. 3. Prolaps ektopické ureterokély u 10denní dívky.

Urethral polyp

Urethral polyp is a fleshy interlabial mass in the urethral meatus. Its color varies between pink and dark red, usually is peduncle-shaped, and can be hemorrhagic to ulcerous. Urethral polyp is considered the pediatric equivalent of a urethral caruncle in postmenopausal women [27]. Chronic inflammation and estrogen deficiency are considered etiological factors. A urethral polyp usually grows from the posterior urethra and may be clinically mistaken for a paraurethral cyst [5]. The incidence of urethral polyps is higher in boys, and they are located in the proximal part of the urethra. Neonatal urethral polyp has been described only in case reports [28,29]. A urethral polyp is usually asymptomatic and may be discovered incidentally during a routine pediatric examination, while sometimes it presents with a protruding mass from a urethral introitus with bleeding, hematuria, and acute urinary retention [27]. The surgical removal of the polyp under cystoscopic control is a preferred treatment approach

with a low risk of recurrence. A case report of a giant urethral polyp measuring 6 × 3 × 3 cm has been described. While expectant management should be considered before surgery, this patient had to be cured by primary surgical excision of the lesion [28].

Vaginal and hymenal polyps

Fibroepithelial polyp of the vagina is a mucosal polypoid lesion with a core of connective tissue covered by benign squamous epithelium. Only a few case reports of vaginal polyps in neonates have been described so far. The treatment is based on the excision of the polyp while the risk of its recurrence is rare [30–32].

A hymenal polyp is an elongated protrusion of hymenal tissue from the hymenal margin. The majority of hymenal polyps are small. They are usually found in the cranial and caudal positions while the lateral position is less frequent. They are of embryonic origin from the urorectal septum. Most hymenal polyps resolve spontaneously and are rarely observed after the age of three [33,34].

Urethral prolapse

Urethral prolapse is a rare lesion that occurs at any age, but with two peaks: in childhood age and postmenopause [22]. The underlying cause remains uncertain but a lack of estrogens is believed to play a role [35]. Urethral prolapse is confirmed upon physical examination presenting as an edematous hemorrhagic tissue arising from and surrounding the urethral meatus, which distinguishes it from other types of interlabial masses. The surface of the mass is covered with urethral mucosa that has prolapsed from the urethral meatus. The position of the vaginal introitus is caudal to the prolapsed urethra, the color of which changes from red to dark blue depending on the degree of vascular supply disorder. Conservative expectant management or surgical treatment with complete or partial reduction of the prolapsed mucosa circumferentially over a urethral catheter should be considered based on the severity of symptoms (mild to serious) [36]. The risk of recurrence is low but the risk of postoperative stenosis of the urethral meatus should be pointed out [35].

Neonatal genital prolapse

Neonatal genital prolapse is a rare condition with prolapse of the uterus and vaginal walls from the vaginal introitus, resistant to a simple reduction. Majority of cases are associated with congenital abnormalities of the spine, such as neural tube defects [37–40], while only a few girls with neonatal genital prolapse have central nervous system malformations. A combination of intrauterine growth restriction with prematurity has a major impact on pelvic floor muscle weakness, which is considered a prerequisite for neonatal genital prolapse. It was first described in a neonate born at gestational week 30 with a birth weight of 800 g [38,41]. Long-term increase of intra-abdominal pressure due to prolonged postnatal ventilation and constipation are considered other risk factors of genital prolapse in preterm infants [38].

Several treatment methods have been proposed based on case reports, ranging from digital reduction to insertion of an intravaginal self-retaining device for pelvic floor support and partial labial fusion. In severe cases, surgery may be performed: ventrosuspension of the uterus, transvaginal slings, and sacral cervicopexy [42].

Duplicated rectum

Duplication of a colon is a congenital anomaly with gastrointestinal symptoms or genitourinary manifestations secondary to the formation of an internal fistula. Rectal duplication may cause neonatal bladder outlet obstruction with hydronephrosis, recurrent urinary tract infections, or a rectovaginal fistula [43]. Rarely, it presents as an interlabial mass in a newborn [43,44].

Discussion

An interlabial mass in a newborn girl is a rare condition, the cause of which can usually be confirmed by physical examination only. The main diagnostic clues include the anatomical location of the mass relative to the vaginal introitus and urethral meatus, as well as the general appearance of the mass. To evaluate the entire contour of the mass and determine important landmarks such as the external urethral meatus and vaginal introitus, wide adduction of the newborn's hip is required. Cystic or solid nature, color, and location of the mass should be noted. The surrounding anatomical structures should also be assessed, including position of the urethral meatus and configuration of the hymen [5]. For example, normal location and appearance of the vaginal introitus and displacement of the urethral meatus are characteristic of a paraurethral cyst [7]. Depending on the nature of the finding, a newborn girl with an interlabial mass should be examined by a pediatrician, gynecologist, surgeon, or urologist.

Congenital vaginal obstruction should be excluded first. Therefore, the newborn girl should be examined for patency of the vaginal canal. Transabdominal ultrasonography should be performed to rule out hydrocolpos due to an imperforate hymen [13]. Hydrocolpos can also be seen with other causes of vaginal obstruction, such as transverse vaginal septum or distal vaginal agenesis [45,46]. However, only in a low positioned transverse vaginal septum, both hydrocolpos and an interlabial mass bulging from a closed vaginal introitus may occur.

The structure of the interlabial formation is crucial for the differential diagnosis, particularly whether it is cystic (e. g., paraurethral or hymenal cyst, imperforate hymen with hydrocolpos, prolapsed ectopic ureterocele) or solid (e. g., urethral or vaginal polyp, rhabdomyosarcoma). Color is another important feature of the interlabial mass. If it is red, prolapse of an ectopic ureterocele

should be suspected, while a yellow color is characteristic of paraurethral and hymenal cysts or hydrocolpos due to an imperforate hymen. A well-focused gynecological examination, including the assessment of appearance and color of the interlabial mass, and its location in relation to the urethral meatus and vaginal introitus may help clinicians to distinguish lesions requiring surgery from those that resolve on their own and need only observation [2].

Majority of interlabial masses are diagnosed after birth, but e. g., hydrocolpos should be found as a cystic mass in the small pelvis already during pregnancy [47]. Rarely, hydrocolpos may be detected prenatally along with oligohydramnios and abnormalities of the anorectum and genitourinary tract [13]. Ovarian cysts, duplication cysts, mesenteric cysts, meconium cyst, and urachal cysts should be considered in the differential diagnosis of prenatally diagnosed intra-abdominal cystic formations [48].

Conclusion

The correct diagnosis of a congenital interlabial mass, based on the knowledge of its various causes and focused postnatal pediatric and gynecological examination, is important for the choice of an appropriate early treatment method or simple observation management and waiting for spontaneous resolution. In particular, early treatment of hydrocolpos and a prolapsed ureterocele causing lower urinary tract obstruction is important.

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Submitted/Doručeno: 18. 6. 2024
Accepted/Přijato: 23. 7. 2024

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Publication ethics: The Editorial Board declares that the manuscript met the ICMJE uniform requirements for biomedical papers.
Publikační etika: Redakční rada potvrzuje, že rukopis práce splnil ICMJE kritéria pro publikace zasílané do biomedicínských časopisů.
Conflict of interests: The authors declare they have no potential conflicts of interest concerning the drugs, products or services used in the study.
Konflikt zájmů: Autoři deklarují, že v souvislosti s předmětem studie/práce nemají žádný konflikt zájmů.