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PELVIC EPIDERMOID CYST IN A TEENAGER: AN EXTREMELY RARE CLINICAL CASE

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Abstract. Epidermoid cysts are defined as benign tumors of the skin that evaluate from the ectodermal germ layer. Typical localization is face, neck, scalp, hands, and much less frequently described unusual locations: brain, gonads, spleen, kidneys. The evolution of cysts of atypical localization is associated with impaired migration of ectoderm cells during embryogenesis. An epidermoid cyst in the child's pelvic is an extremely rare clinical case. In the available literature, only two descriptions of epidermal cysts of pelvic localization among children are found. The clinical course of epidermoid cysts can be asymptomatic or accompanied by pain, lower urinary tract symptoms, disruption of internal organs. Indications for surgical treatment of such cysts are their possible inflammation, compression of neighboring organs with disruption of their function, and an extremely low but probable risk of malignancy. We present a clinical case of successful minimally invasive treatment of a 17-year-old boy with a pelvic epidermoid cyst, suffering from long-term abdominal pain syndrome.

Keywords: *epidermal cyst, pelvic cyst, cysts in children, extraorgan cyst, pediatric urology, laparoscopy, surgical treatment*

ЭПИДЕРМАЛЬНАЯ КИСТА МАЛОГО ТАЗА У ПОДРОСТКА: ЭКСТРЕМАЛЬНО РЕДКИЙ КЛИНИЧЕСКИЙ СЛУЧАЙ

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Резюме. Эпидермальные кисты определяют как доброкачественные опухоли кожи, происходящие из эктодермального зародышевого листка. Типично они локализуются на лице, шее, волосистой части головы, кистях, значительно реже встречаются необычные локализации: головной мозг, гонады, селезенка, почки. Происхождение кист нетипичной локализации связано с нарушением миграции клеток эктодермы в процессе эмбриогенеза. Эпидермальная киста полости малого таза у ребенка — экстремально редкий клинический случай. В доступной литературе нами найдено всего два описания эпидермальных кист тазовой локализации у детей. Клиническое течение эпидермальных кист может быть как бессимптомным, так и сопровождаться болевым синдромом, нарушением мочеиспускания, нарушениями работы внутренних органов. Показаниями к хирургическому лечению таких кист являются их возможное воспаление,

сдавление соседних органов с нарушением функции и крайне низкий, но вероятный риск малигнизации. Нами представлен клинический случай успешного малоинвазивного лечения мальчика 17 лет с эпидермальной кистой малого таза, страдающего длительным абдоминальным болевым синдромом.

Ключевые слова: *эпидермальная киста, киста малого таза, кисты у детей, внеорганная киста, детская урология, лапароскопия, хирургическое лечение*

INTRODUCTION

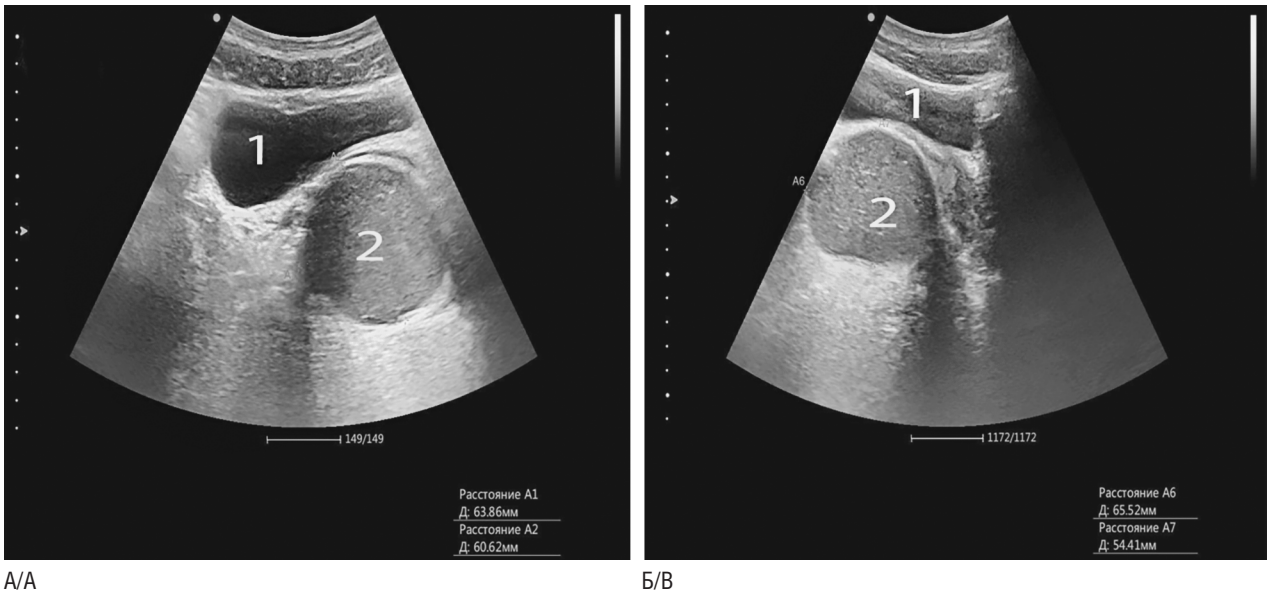
Epidermoid cysts (epidermal inclusion cysts) are defined as benign skin tumors developing from the ectodermal germinal sheet [1]. Most commonly, this pathology develops in individuals aged 19–45 years and usually the cysts are located on the face, scalp, neck, hand, and foot [1–3]. Unusual localizations such as brain, gonads, bones, spleen, kidney and other internal organs are much less common [4]. Usually, cysts do not bother patients and are discovered incidentally, but when they are large, they may compress neighboring organs, lead to lower urinary tract obstruction, pain syndrome or cause discomfort to patients [2, 4, 5]. The available literature describes two cases of treatment of children with pelvic epidermoid cysts. Thus, this type of localization is extremely rare in paediatric practice [3, 6].

CLINICAL CASE

Patient E., 17 years old, was admitted to the department of paediatric urology of St. Petersburg State Paediatric Medical University on 23 January 2023 with complaints of recurrent abdominal pain. According to the anamnesis: during the last year he was examined in a hospital at the place of residence due to painful abdominal syndrome, further ultrasound examination (USG) revealed a voluminous cystic formation in the small pelvis. After discharge he was consulted by a paediatric urologist-andrologist and sent for additional examination to the clinic of the Pediatric University.

Ultrasound: a rounded formation with thin walls was detected in the small pelvis, the formation was deforming the bladder, the content was of medium echogenicity, inhomogeneous, with hyperechogenic inclusions (Fig. 1).

Uroflowmetry: the curve is flattened, micturition volume — 400 ml, maximum flow rate — 17.4 ml/s, average flow rate — 7.8 ml/s, residual volume — 8 ml. Magnetic resonance imaging (MRI): the formation is located in the pelvis, up to 6.5 cm in size, to the left of the prostate gland, partially deforms the left lobe of the prostate, the content is fluid with inclusions (Fig. 2).



А/А Б/В

Fig. 1. Pelvis ultrasound: A — in the frontal plane; B — in the sagittal plane. 1 — bladder; 2 — epidermoid cyst

Рис. 1. УЗИ малого таза: А — во фронтальной плоскости; Б — в сагиттальной плоскости. 1 — мочевой пузырь; 2 — эпидермальная киста

Blood test: alpha-fetoprotein — 1.22 IU/ml (norm: 0.00–15.00 IU/ml), LDH — 157 units/L (norm: 125.00–220.00 units/l), beta-HCG — 1.3 IU/ml. The child was consulted by a paediatric oncologist — removal of the cystic neoplasm was recommended.

Intraoperative cystourethroscopy was performed to exclude the connection of the lower urinary tract with the cyst cavity, during which no signs of communication were detected. The bladder wall showed no visible changes. Laparoscopic trocars (5 mm) were placed in the periapical, left and right iliac regions. The peritoneum was dissected in the bladder apex, after which a tumor-like formation was identified and mobilized in the retrovesical space. The cyst was completely isolated and dissected, after which it was evacuated from the abdominal cavity. Doughy, granular, cream-colored contents were extracted at autopsy (Fig. 3, A).

The postoperative period was uncomplicated. The patient received antibacterial, symptomatic, and external therapy. The urethral catheter was removed on the 3rd day. Control uroflowmetry was performed on the 7th day after the operation. It showed an increase in the average flow rate from 7.8 ml/s to 9.3 ml/s, maximum flow rate from 17.4 ml/s to 20.7 ml/s with a micturition volume of 410 ml.

Tissue diagnostics: the cyst wall consists of fibrous connective tissue, with numerous dilated blood vessels, small focal hemorrhages. The cyst lining was formed by multilayer squamous keratinizing epithelium, which corresponds to the pathological picture of an epidermal cyst (Fig. 3, B). On the 7th day after surgery, the child was discharged.

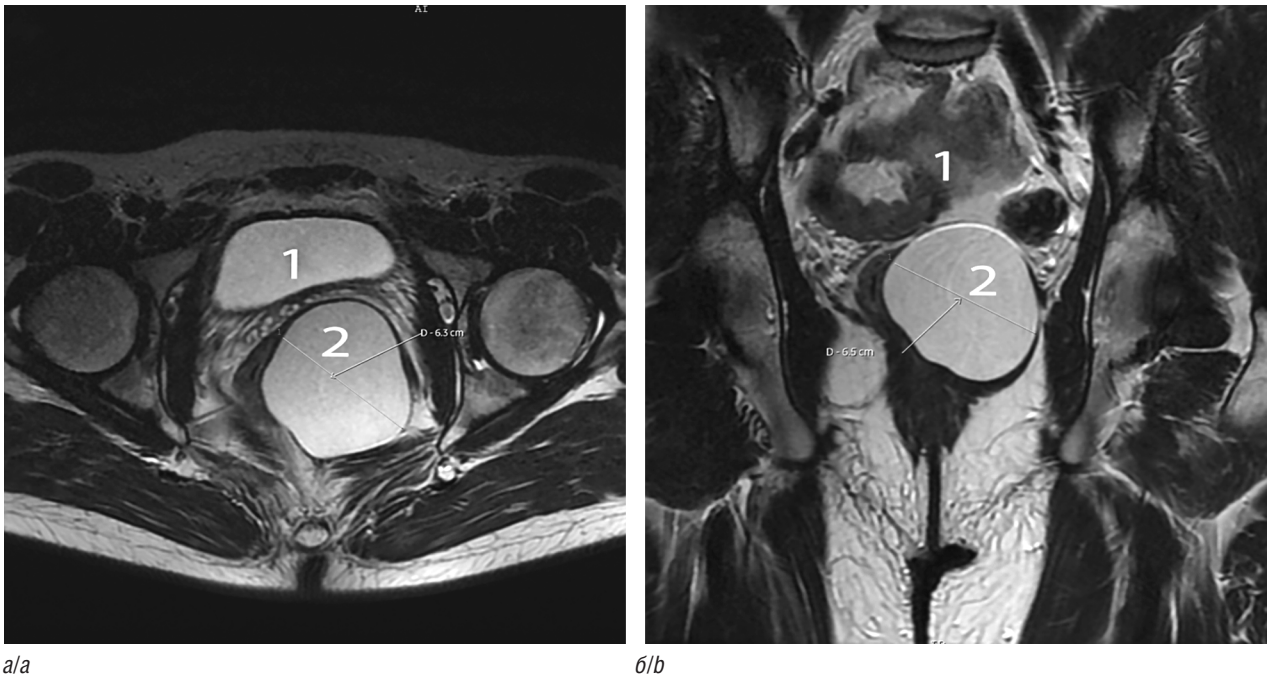
Next time, the boy was examined in the department after 1 year. There were no complaints of abdominal pain during this period, the patient feels healthy. There are no signs of new cystic formations in the pelvic cavity at ultrasound. Uroflowmetry curve was slightly flattened, average flow was 9.9 ml/s, maximum flow was 18.5 ml/s with a volume of 250 ml.

DISCUSSION

Epidermoid cysts are benign tumors of ectodermal origin. This pathology was first described by M.B. Dockerty and J.T. Pristley (1942) and defined as a cystic formation of unclear etiology [7].

A number of authors believe that the origin of congenital epidermal cysts is associated with embryonic implantation of ectoderm cells [2, 4, 5]. B. Fakhir et al. (2009) reported that acquired epidermal cysts can occur due to trauma or undergone surgery [5].

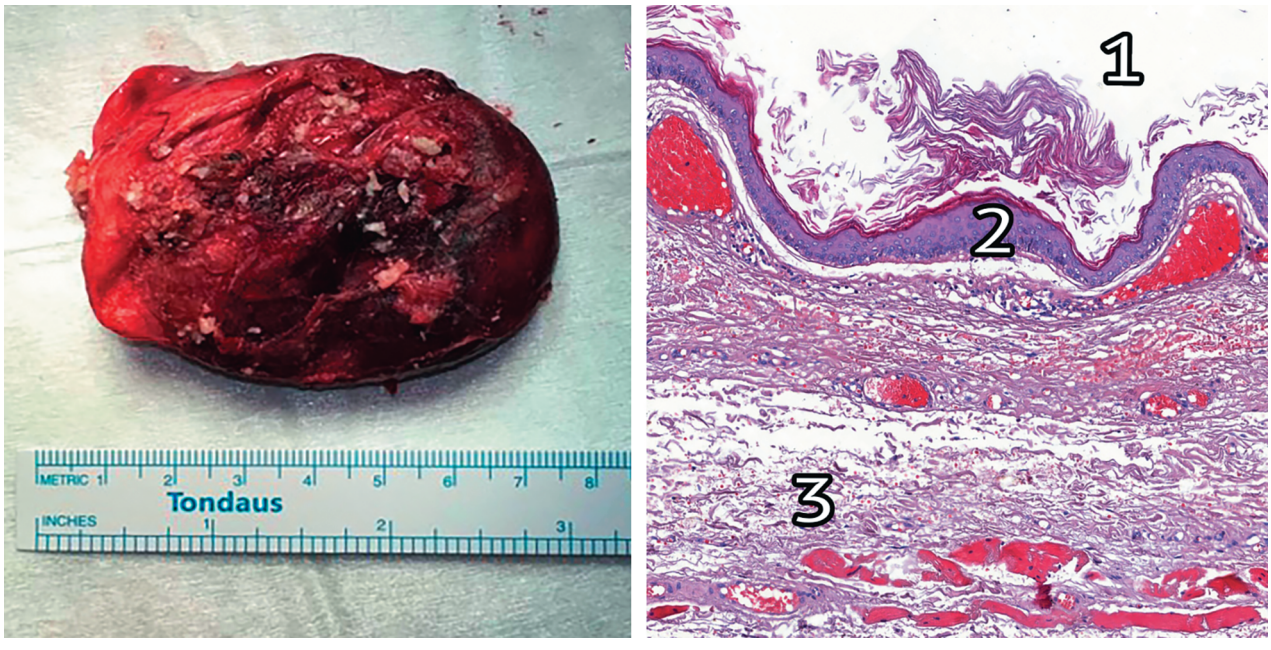
The typical localization for this pathology is the face, scalp, neck, chest and genital skin (scrotum,



a/a *б/b*

Fig. 2. MRI: *a* — frontal plane; *b* — sagittal plane. 1 — bladder; 2 — epidermoid cyst

Рис. 2. МРТ: *а* — фронтальная плоскость; *б* — сагиттальная плоскость. 1 — мочевой пузырь; 2 — эпидермальная киста



а/a *б/b*

Fig. 3. Removed cyst (*a*); histological specimen (*b*). 1 — cyst cavity; 2 — epithelial lining; 3 — fibrous connective tissue

Рис. 3. Удаленная киста (*а*); гистологический препарат (*б*). 1 — полость кисты; 2 — эпителиальная выстилка; 3 — волокнистая соединительная ткань

penis) [1, 2]. Unusual localizations such as the brain, gonads, bones, spleen, kidneys and other internal organs are described much less frequently [2, 3, 7, 8].

Epidermoid cyst located in the pelvis, which is not associated with internal organs, is an extremely rare clinical case. We found two descriptions of pelvic epidermoid cyst in a child in the available literature [3, 6]. Meanwhile, F.Z. Fdili Alaou et al. (2012) reported 15 cases of epidermoid cysts in adults [4].

N.S. Hyseni et al. (2009) believe that clinical manifestations depend on the compressive effect on organs and tissues. The patient described in their work suffered from abdominal bloating and pain [3]. In the second case presented by Y. Kato et al. (1998), the pathology was asymptomatic [6]. A number of authors also describe the development of pain syndrome, urinary disturbance due to urethral compression in adults [2, 5, 9]. In our clinical case, the patient was bothered by recurrent abdominal pain.

Large cysts can be palpated through the anterior abdominal wall, and sometimes the cyst can be palpated rectally [10]. In our clinical case, the cyst was not detected rectally.

Since epidermoid cysts are usually benign tumors, specific markers of malignant growth are informative only in cases of neoplastic transformation [2].

According to J. Pritesh et al. Pritesh et al. (2018), it is possible to visualize a delimited rounded formation of mixed echogenicity in ultrasonography [2]. F.Z. Fdili Alaoui et al. (2012) report that such formations in women can be taken for ovarian cysts [4].

According to a number of authors, computed tomography (CT) scans show the absence of a homogeneous fluid component in these cysts, which distinguishes them from other common cystic formations such as lipomas, fibromas, and desmoid tumors [2, 5, 8]. For a more accurate diagnosis, it is better to use MRI, in which it is possible to see a clearly delineated formation with a heterogeneous signal intensity in both T1- and T2-modes [4, 5, 10]. According to B. Fakhir et al. (2009), contrast accumulation by the surrounding tissues will allow to assess the nature of the cystic formation, as well as to predict the possibility and degree of mass effect [5].

According to most authors, histological examination demonstrates that an epidermal cyst is a cavity with fibrous connective tissue walls containing full-blooded vessels. The cyst lining consists of multilayered squamous keratinizing epithelium, and horny scales may be detected

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in the lumen [2, 4, 10]. Our sample demonstrates the same histological picture.

The important differential sign distinguishing epidermal cysts from other cysts is the absence of skin appendages (sebaceous and sweat glands, hair follicles) [1]. Tumor diagnostics of the removed cyst showed no skin appendages in the cyst lumen.

Malignisation of such formations is considered improbable [1, 2, 4]. M.V. Kuritsyn (2006) describes 4 cases of malignant neoplasms formed from the epithelial lining of epidermoid cysts in 50–58-year-old women [11]. It is known that the proportion of children with neoplasms of various localizations can reach 2.3% [12] and that the main problem of paediatric oncology remains late diagnosis [13].

Oncological vigilance is one of the main directions of the diagnostic process against the background of increasing cancer morbidity rates in St. Petersburg, which is associated, among other things, with improving the quality of examination and increasing the vigilance of physicians with regard to cancer [14].

CONCLUSION

Pelvic epidermoid cyst in a child can manifest with prolonged abdominal pain syndrome. Surgical removal of the formation with subsequent histological examination is the only method of definitive diagnosis and treatment of this extremely rare pathology.

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