

Intraparenchymal Epididymal Cyst (IEC) 4 cm in Diameter in a 15-Year Old Male Patient; a Case Report and Review of the Literature

Dimitrios Patoulias, Maria Kalogirou, Ioannis Patoulias*

1st Department of Pediatric Surgery, Aristotle University of Thessaloniki, GH G. Gennimatas, Thessaloniki, Greece

* Corresponding author: M. Alexandrou 3B, Peuka, Thessaloniki, Postal code 57010; e-mail: dipatoulias@gmail.com

Summary: Intraparenchymal epididymal cysts (IECs) are benign cystic formations of the epididymis of unknown pathogenesis, which typically appear in adolescence or adulthood. In patients older than 14 years old their prevalence is doubled. After systematic and thorough research of the current literature, we did not find another case report of intraparenchymal epididymal cyst with similar dimensions. The male patient, 15 years old, visited our outpatient department complaining of pain in the right hemiscrotum. Diagnosis of IEC was confirmed after the conduction of ultrasound examination. Patient underwent surgical exploration of the right hemiscrotum. Resection of the IEC followed. Postoperative course was uneventful, with recession of the symptoms. In our opinion, IECs should be surgically removed, either when they are symptomatic or when they are asymptomatic, but of a diameter greater than 1 cm and without regression tendency.

Keywords: *Intraparenchymal epididymal cyst (IEC); Epididymis; Painful hemiscrotum*

Introduction

Intraparenchymal epididymal cysts (IECs) are benign, single spaced, containing serous fluid, formations, lined by cuboidal or columnar epithelium (1, 2). Accidental identification of an IEC occurs in 0.8% of all adolescents that undergo ultrasound examination of the scrotum (1). Nidzielski J. et al. (1) studied 45 cases of adolescents with IECs, noting that in 75–80% they were solitary, in 20% there were 2 cysts and in 5% there were 3 or more cysts, unilateral or bilateral. It is in fact an extratesticular cystic lesion, similar to spermatocele, regarding to the composition of the fluid and the histological structure of its capsule. They differ in: a) the localization, because spermatocele originates from the head of the epididymis, while IEC can originate from every region of the epididymis and b) the composition of the containing fluid. In spermatocele it consists of sperm cells, lymphocytes and cellular debris. Fluid cannot be serous, but milky as well (3).

Aim of this study is to present a case of an adolescent with an IEC 4 cm in diameter – especially after noticing that there are no case reports of IECs of such a diameter in current literature – and our proposal on the appropriate diagnostic and therapeutic approach.

Case report

A 15 years old male patient with free medical history visited our outpatient department complaining of the presence of a lump that he first palpated in the right hemiscrotum

12 months ago during self-examination. He reported mild pain, exacerbating in the context of physical activity. During inspection of the scrotum there were no signs of inflammation, but the cyst protruded as an intrascrotal mass above the right testicle. Further physical examination revealed the presence of a sizeable, painless, fluctuating lesion in the upper right hemiscrotum, clearly distinguished from the ipsilateral testis. Transillumination was indicative of a rather cystic lesion. Right testicle was orthotopic, painless when palpated, while it was impossible to distinguish the boundaries between the epididymis and the lesion. No other



Fig. 1: IEC 4 cm in diameter, which occupies the head and the body of the epididymis.

pathological manifestations were detected at the rest of the scrotum and groin.

A single spaced, thin walled, cystic lesion, within the parenchyma of the head and the body of the right epididymis, 4 cm in diameter was depicted in the ultrasound. The two testes had normal imaging features, with dimensions $26 \times 17 \times 14.5$ mm the right and $25.8 \times 17.2 \times 13.8$ mm the left. Imaging evaluation of the rest scrotal structures, the groin and the urinary tract was normal.

Elective surgical procedure under endotracheal anesthesia followed. Access was achieved by a transverse incision in the middle of the right hemiscrotum, followed by the transection of the dartos, the external spermatic fascia, the cremasteric fascia, the internal spermatic fascia, the parietal and visceral tunica vaginalis and the externalization of the testis and the epididymis. IEC on the head and the body of the epididymis was identified. Careful enucleation of the cyst followed (Figure 1).

Epididymal capsule was sutured continuously by using Vigryl rapid 6/0. Testis and epididymis were repositioned in the ipsilateral hemiscrotum, followed by the closure of the surgical wound in the anatomical order.

Results

Postoperative period was uncomplicated and the patient was discharged home on the first postoperative day. Histopathological report described a cystic lesion with smooth transparent wall and a transitional epithelial lining, containing serous fluid, but no spermatozoa. Three years later, after a thorough follow-up on a 6-month basis, patient is free of any complication or relapse or accompanying correlating signs.

Discussion

IEC is often identified in patients with cryptorchidism, cystic fibrosis, Von Hippel-Lindau disease, polycystic kidney disease, as well after prenatal exposure to diethylstilbestrol (2, 4, 5). The incidence of IECs doubles after the age of 14 years (1, 6). Sempere CFS et al. (7) studied cases of 15 boys with IECs aged 1–16 years old (average 11.5 years). Left varicocele co-existed with IEC in 40% of these patients (6/15), while 26% of the patients (4/15) had a medical history of orcheoepididymitis. Finally, in half of these cases, diagnosis of an IEC was made accidentally, while the patient underwent physical examination for another reason. Thus, they emphasize on the need of thorough diagnostic approach in patients with IEC, in order to confirm or exclude the presence of varicocele.

Differential diagnosis should include intrascrotal cystic lesions, which are divided – based on their localization – into intratesticular (testicular cyst, tunica albuginea cyst, epidermoid cyst) and extratesticular (epididymal cystic lymphangioma, spermatocele, paratesticular abscess, epididymal adenomatoid tumor, varicocele) (3). Absence of spermatozoa in the cyst's fluid and its attachment to the head

and the body of the epididymis contributed to the exclusion of spermatocele in our case (8).

Regarding to the pathogenesis of IECs the following cited might play a significant role: a) hormonal disorders stemming from toxic agents acting either prenatally (such as diethylstilbestrol or Cannabis indica) or after birth (9, 10, 11), b) the degenerative process and c) the existence of obliteration or stenosis of the epididymal ducts with subsequent pro-stenotic dilatation. The latter evolves progressively, leading to the development of a cystic lesion, which possibly exerts pressure on the adjacent seminiferous tubules (3). This etiologic approach is strengthened by the observation of Blau et al. (12) on the incidence of IECs in patients with cystic fibrosis. Cystic fibrosis is associated with the absence of the vas deferens in 90% of all cases. Based on this etiologic approach, the peripheral obstruction of the secretory system leads to the cystic dilatation of the epididymal secretory system and finally the development of an IEC.

Sharpe RM et al. (13) and Skakkebaek NE et al. (14) consider as possible the correlation between the IECs and the testicular dysgenesis syndrome. Wollin M. et al. (15) believe that the IEC results from the progressive dilatation of the vestigial remnants, which do not communicate with the secretory epididymal ducts. If it is considered as a result of cystic dysplasia of the affected epididymis, with the impaired development of the mesonephric duct being the embryological substrate, then preventive ultrasound examination of the kidneys is absolutely indicated. In our case, we conducted also an ultrasound scan of the kidneys during the preoperative period (16). Belet U. et al. (17) emphasize on the necessity of the exclusion of autosomal inherited polycystic kidney disease, which can co-exist with IEC.

In our case, IEC was 4 cm in diameter, finding that suggests that lesion's dimensions may be greater than those reported so far in the literature. In our case, it is possible that the size of IEC gradually increased, however, due to the patient's age, parents could not have noticed the scrotal mass. Erikci V. et al. (18) reported 42 patients with IEC (22 left, 16 right and 4 bilateral), aged from 2 months to 16 years old (average 10.7 years). Cysts' diameter ranged from 2 to 20 mm. Hegazy AF. et al. (11) studied 20 boys aged from 3 months to 15 years old (average 11.5 years) with IEC from 0.3 to 3 cm in diameter. Pieri S. et al. (19) treated 25 patients with symptomatic IEC, 5 cm or greater in diameter. However, all the patients that underwent treatment were adults.

Ameli M. et al. (20) reported a case of a torsion of an epididymal cyst ($4 \times 4 \times 3$ cm) in a 14-year old boy. However, it was a case of appendiceal torsion (embryonic remnant of epididymis) and not a case of an IEC.

Homayoon K. et al. (2) studied 20 patients with identified IEC. Patient's average age at diagnosis was 10.5 years. 15 patients presented complaining of a palpable scrotal mass, while 4 patients suffered from scrotal pain. Cysts' diameter ranged from 3 to 30 mm.

The context of treatment of an IEC is determined by the cyst's diameter, the symptomatology and the development of

complications (3, 6, 21). 51.8–75% of the cysts are asymptomatic, found accidentally during physical examination, while 25–49.2% of them are symptomatic (2, 6) and 50% of them regress automatically within the next 3–35 months (average 18 months) (2). Conservative approach should be selected in patients with asymptomatic IEC, less than 1 cm in diameter. On the other hand, elective surgical removal should be conducted when: a) the IEC is asymptomatic, greater than 1 cm, without regression tendency after 12 month surveillance (18), b) the IEC is symptomatic, regardless of its diameter and c) there are signs and symptomatology of acute scrotum due to inflammation, intracystic bleeding or secondary epididymal torsion. In our case, surgical treatment was absolutely indicated, due to the presence of persistent symptoms for at least 4 months and the large diameter of the cyst, greater than all those that are described in patients of the same age in literature. Indicated method of treatment in childhood is the open surgical approach through scrotal incision, either transverse or on the scrotal raphe. Operative strategies in the treatment of IECs and spermatocele in young adults are almost the same. Kauffman EC et al. (22) and Cagiu C. et al. (23) attempted the microscopic assisted spermatocelectomy in order to prevent accidental epididymal injury. It is possible that the microscopic assisted resection of IECs will be the preferable treatment option in the future. Hegazy AF et al. (11) and Pieri S et al. (19) described a conservative technique, consisting of puncture of the IEC, aspiration of its fluid and injection of polidocanol. This technique has been attempted in adults, but not in childhood yet (11).

Despite the thorough approach and resection of the IEC in our case, the possibility of accidental epididymal injury because of immediate vicinity of the efferent epididymal tubules, which could lead to infertility in the future, cannot be excluded. Based on this hypothesis, we believe that a periodic follow-up of the patient and the conduction of spermogram in a reasonable time are required. Besides, according to the clinical study conducted by Weatherly D. et al. (24), who encountered 91 men, IECs are not related to infertility ($\chi^2_{(df=1)} = 0.362$, $p = 0.55$).

Conflict of interest

None of the contributing authors have any conflict of interest, including specific financial interests or relationships and affiliations relevant to the subject matter or materials discussed in the manuscript.

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