



Epidermoid inclusion cyst after urethroplasty: A rare complication

Volkan Sarper Erikci¹,  Merve Dilara Oney¹, Gokhan Koyluoglu²

¹Departments of Pediatric Surgery, University of Health Sciences, Tepecik Training Hospital, Izmir, Turkey

²Departments of Pediatric Surgery, Katip Celebi University, Tepecik Training Hospital, Izmir, Turkey

ABSTRACT

Epidermoid inclusion cysts (EIC) is a disease caused by the nidation of the epidermal tissues and sebaceous materials into the dermal and subcutaneous layers. This entity is usually observed after trauma and surgical procedures. A 5-year-old boy with a ventral penile mass was admitted to our department. The history of the patient revealed that he was operated at the age of 1 year for hypospadias in a different medical center. The patient's history also revealed that during the initial operation, a skin graft was used for covering urethroplasty. One year later the operation, a ventral penile mass was observed by his mother during wash. The mass continued to increase in size reaching of 1 cm in diameter. The mass was excised and the histopathological study revealed that the mass was compatible with an EIC. With 2 years of follow up the patient is disease free. EICs after penile surgical interventions may be a factor of anxiety for both the children and their parents. In order to avoid this unwanted complication, implantation of the superficial tissue layers into the deep the dermal and subcutaneous tissues should be avoided.

Key Words: Epidermoid inclusion cysts, hypospadias, complication, children.

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✉ Volkan Sarper Erikci, MD, Associate Prof.
Departments of Pediatric Surgery, University of Health
Sciences, Tepecik Training Hospital, Izmir, Turkey
E mail: verikci@gmail.com

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Introduction

Epidermoid inclusion cysts (EIC) are rare in children. The pathophysiological mechanism in this disease is postulated to be the implantation of superficial tissue layers including the epidermal cells and sebaceous glands into the deep dermal and subcutaneous

tissues [1, 2]. The typical occurrence is a painless swelling at the affected body sites. These cysts can occur anywhere on the body and typically present as nodules directly underneath the child's skin. They are usually mobile and the size of these cysts can be variable ranging from a few millimeters to several centimeters in diameter. These lesions may remain stable or progressively enlarge over time. These cystic masses usually become painful to the patient and may present as a fluctuant filled nodule below the patient's skin. Herein, after an unsuccessful surgical intervention for hypospadias, a 5-year-old boy

with an EIC located at the ventral aspect of the penis is presented and the topic is discussed and a brief literature review is given.

Case report

A 5-year-old boy complained of a penile mass located at the ventral aspect of the penis. The history of the patient revealed that he had been operated for hypospadias at the age of 1 year. At the initial operation a skin graft had been used in order to cover urethroplasty suture line. The family of the patient declared that initially the mass was small, 1 year after the operation it started to grow rapidly (Fig. 1).



Fig. 1. Preoperative appearance of penile EIC.

Under general anesthesia a surgical intervention was performed with a vertical incision on the ventral aspect of the penis. The mass with a dimension of 2x1.5x0.7 cm was totally excised (Fig. 2, 3). The urethra was not catheterized intra-operatively and the fashion of penile skin closure after EIC removal was interrupted simple suture using 5/0 polyglycolic acid. Histopathological examination findings were compatible with epidermal inclusion cyst with a capsule at the outermost and inside of which there was keratinized material. With an uneventful

postoperative follow-up of 2 years, he is disease free without any symptoms of recurrence.



Fig. 2. Operative view of EIC during excision.



Fig. 3. Postoperative view of the penis after removal of EIC.

Discussion

As an intriguing clinical entity, EIC is supposed to occur due to implantation of the superficial squamous cells and oleaginous materials into deep tissues following an injury and surgical procedure [1, 2]. Accepted as real cysts, these masses contain keratinized material inside and have an outer capsule composed of keratinized squamous epithelial cells. EICs may either be congenital or acquired in origin. Congenital forms of penile EIC have been supposed to result from

abnormal embryologic closure of the median raphe [3]. Penile surgery and trauma are two of these factors. It has been suggested that during penile surgical interventions like circumcision or hypospadias surgery, epidermal cells are embedded within the dermis [4,5]. In some of these cases no initiating factor for EIC development is present and these are called as idiopathic forms of penile EIC [6].

EICs may be single or multiple and the size of the mass varies. Accumulation of epidermal materials including secretions and debris leads to formation of a cyst. This painless swelling gradually increases as the time passes [7]. In accordance with those reported previously, the cystic mass in the presented case was initially small, but later it became larger rapidly. Physical examination is enough in diagnosing these masses. In doubtful cases imaging modalities such as ultrasonography and other means of radiological studies may be useful in conforming the diagnosis. There are various diseases in the differential diagnosis of penile EICs and these are diverticula or fistulae at urethra, dermoids or teratomas [3]. Physical examination findings in our case was enough to diagnose EIC and no other diagnostic imaging studies were performed.

There are various complications that can be observed during the surgical intervention for EIC. These are rupture of the cyst, infection, hematoma and rarely carcinomas [8,9]. Rupture deserves special attention because rupture of the cyst almost always ends up with an inflammatory reaction due to release of keratin inside the cyst. The time interval in our case is relatively long with a time period of 4 years between the initial hypospadias surgery and surgical excision of the EIC. Hopefully, despite rather long delay for treatment of EIC in our case, no complication was observed in our case during pre- and postoperative period.

Nevertheless, once EIC is diagnosed at the penis, there should not be delay in the management of these masses and gentle surgical excision should be the choice in order to avoid above mentioned complications.

Complete surgical excision is curative in the management of penile EIC. In order to avoid local recurrences, meticulous dissection of the mass is paramount [1]. One of the important issues in the management of these masses is that there should not be violence against to the capsule of the EIC during surgical excision. Otherwise, local seeding of the surface materials into the dermal subcutaneous tissues may lead to local recurrence.

Histopathological examination is the cornerstone in confirming the diagnosis of EIC. These masses are typically lined by keratinized squamous epithelium and contain sebaceous material inside [7]. Histopathological examination findings were compatible with EIC containing cheesy material inside in our patient. The patient is disease free after a 2 year-follow up period.

Penile EICs are rare during childhood and can pose diagnostic dilemmas for health providers. Timely and appropriate surgical treatment is important in preventing complications such as infection, hematoma and rarely carcinomas. Most of the reported cases of EICs are children with penile masses after circumcision [10,11]. As a cause of urethro-cutaneous fistula, a penile EIC following urethroplasty has also been reported [3]. After an English language literature search for complications following hypospadias surgery in children, we did not find out EIC following hypospadias surgery in childhood. To our knowledge, after hypospadias surgery, our case is unique and probably the first case of EIC having an isolated EIC without any other urological complication.

Conclusions

Surgical interventions like circumcision and hypospadias repairs should be performed carefully. This is important in avoiding psychological and surgical trauma that the child may face following penile surgical interventions. During surgery, it is also important to avoid seeding of superficial tissue materials into the dermis and subcutaneous tissue. The diagnosis of EIC in children with penile cystic masses should be kept in mind and a gentle management including total excision of EIC without violating the capsule should be performed.

Compliance with ethical statements

Conflicts of Interest: None.

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ORCID ID of Authors

Volkan Sarper Erikci /0000-0002-9384-2578

Merve Dilara Oney / 0000-0002-2895-2578

Gokhan Koyluoglu / 0000-0002-1140-169X

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