A Rare Complication after Urethroplasty: Epidermoid Inclusion Cyst

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Abstract
Epidermoid Inclusion Cysts (EIC) occur as a result of the implantation of the epidermal keratinized squamous epithelial cells and sebaceous glands into the dermis and subcutaneous tissue after trauma and surgical interventions. A 5-year-old boy with a penile EIC who was operated elsewhere at the age of 1 year for an anterior hypospadias with a usage of skin graft covering urethroplasty is presented and discussed with regard to the foregoing literature. In order to avoid psychological and surgical trauma that can be seen after circumcision and hypospadias surgery in children, all the surgical interventions should be performed carefully and during surgical intervention implantation of the epidermis into the dermis and subcutaneous tissue should be avoided.

Introduction
Epidermoid Inclusion Cysts (EIC) occur as a result of the implantation of the epidermal keratinized squamous epithelial cells and sebaceous glands into the dermis and subcutaneous tissue [1,2]. These masses typically present as painless swellings located at the related locations of body. A 5-year-old boy with a penile EIC who was operated for hypospadias with a usage of skin graft covering urethroplasty is presented and discussed in the light of relevant literature.

Case Presentation
A 5-year-old boy was admitted to our clinic with a diagnosis of penile EIC. The patient had been operated at the age of 1 year for an anterior hypospadias with a usage of skin graft covering urethroplasty. Initially, the mass was reported to be small, but later it started to grow rapidly (Figure 1). Under general anesthesia with a vertical incision on the ventral aspect of the penis the cystic mass was totally excised (Figure 2 and 3). Histopathological examination revealed an epidermal inclusion cyst with a dimension of 2 cm × 1.5 cm × 0.7 cm. The cyst had a capsule at the outer surface and contained keratinized material inside. Postoperative follow-up was uneventful.

Discussion
EICs occur as a result of the implantation of the epidermal keratinized squamous epithelial cells and sebaceous glands into the dermis and subcutaneous tissue after trauma and surgical interventions [1,2]. These masses are real cysts containing keratinized material and are surrounded by keratinized squamous epithelial cells. They can be congenital or acquired. Abnormal embryologic closure of the median raphe is postulated to represent congenital forms of penile EIC [3]. In terms of acquired etiological factors different theories have been proposed for these masses including penile surgery and trauma. It is stated that epidermal cells are implanted within a circumscribed space of the dermis during penile surgical interventions such as circumcision or hypospadias surgery [2,4,5]. Idiopathic forms of penile EIC have also been described [6]. These masses can be single or multiple with variable size. Accumulation of epidermal desquamations, secretions and debris in a closed space leads to formation of a cystic and often painless swelling that gradually increases in size over time [7]. The cystic mass located at the ventral aspect of the penis in the presented case was initially reported to be small, but later the family of the child stated that it started to grow rapidly. Physical examination is all that is needed to diagnose these masses. In doubtful circumstances ultrasonography and means of other radiological evaluations may be used to confirm the diagnosis. The differential diagnosis of penile EICs include urethral diverticula, urethrocysticuteous fistula, dermoid cysts or teratoma. Concerning our case, physical examination of penis was enough to diagnose EIC and no other diagnostic radiological modalities were used. Complications of EIC have been reported including rupture and release of keratin that leads to inflammatory reaction,
infection, hematoma and rarely carcinomas [8,9]. With regard to our patient, although there is a relatively long time period (4 years) between the initial hypospadias surgery and surgical excision of penile EIC, we did not observe any complications in our case during pre- and postoperative period. Nevertheless, once EIC has been detected at the penis following trauma or other surgical procedures, surgical excision of the mass should be a matter of necessity rather than of choice to avoid above mentioned complications. The management of penile EIC is complete surgical excision. Meticulous dissection of the mass is necessary to avoid local recurrences. The capsule of the EIC should not be violated during surgical excision of the mass otherwise local implantation of the epidermal keratinized squamous epithelial cells and sebaceous glands into the dermis may lead to recurrence during follow-up period. Histopathological examination of the excised mass is necessary to confirm the diagnosis of EIC. Typically these cystic masses are lined by keratinized stratified squamous epithelium containing keratinized sebaceous material inside. Histopathological examination revealed an EIC with presence of cheesy material at the cut surface of the mass in our patient. There is no recurrence at 3 months follow-up.

During childhood period, penile EICs are rarely seen and can mimic other disease states. Surgical treatment, if performed early, can prevent complications such as infection, hematoma and rarely carcinomas. Literature on this subject reveals that most patients with EIC were reported following circumcision [10,11]. An EIC of the penis after urethroplasty causing an urethro-cutaneous fistula has also been reported. To our knowledge after hypospadias surgery, there is no pediatric patient with an EIC not associated with an urethro-cutaneous fistula. Presented child is probably the first case of EIC following hypospadias surgery not associated with other complications of hypospadias. In order to avoid psychological and surgical trauma that can be seen after circumcision and hypospadias surgery in children, all the surgical interventions should be performed carefully. During surgical intervention implantation of the epidermis into the dermis and subcutaneous tissue should also be avoided. The possibility of this diagnosis should be kept in mind for the patients with penile cystic masses and managed accordingly.

References