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ONLINE CASE REPORT

Trichilemmal Cyst of the Penis in a Paediatric Patient

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كيسة في غمد جذر الشعرة في قضيب طفل

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ABSTRACT: Paediatric penile cysts are uncommon. We report a five-year-old child with an asymptomatic progressively growing cyst on the ventral aspect of the penis after a hypospadias repair. The patient presented to the Cooper Health Clinic, Dubai, United Arab Emirates, in March 2012. A complete excision of the cyst was performed. Histology results delineated a capsulated benign trichilemmal cyst. No recurrence or complications were reported in the 26 months following the excision. We recommend an early and complete excision of all penile cysts to prevent the risk of urethral obstruction, infection, inflammation and rare malignant changes. This is the first reported case of a penile trichilemmal cyst in a child.

Keywords: Trichilemmal Cyst; Penis; Hypospadias; Child; Case Report; United Arab Emirates.

الملخص: يعد وجود كيسات في القضيب أمرا غير شائع عند الأطفال. وهنا نسجل حالة طفل عمره خمس سنوات كان يشكو من كيسة كانت تنمو بصورة مترقية دون أعراض على الجانب البطني للقضيب بعد عملية تعديل مبال تحتاني. وأحضر المريض لمستشفى كزبر هيلث بدبي في مارس 2012م. وتم استئصال كامل للكيسة. وأثبتت نتائج الفحص الهيستلوجي كيسة حميدة ذات محفظة في غمد جذر الشعرة. وبعد مرور 26 شهرا بعد الاستئصال لم تحدث انتكاسة أو مضاعفات للطفل. إننا ننصح بإجراء استئصال مبكر وكامل لكل كيسة تظهر في القضيب لمنع اختطار انسداد الإحليل، والعدوى والالتهاب، وأيضا التغيرات الخبيثة في حالات نادرة. هذه هي الحالة الأولى المسجلة لطفل مصاب بكيسة في غمد جذر الشعرة في القضيب.

مفتاح الكلمات: كيسة في غمد جذر الشعرة؛ قضيب؛ مبال تحتاني؛ طفل؛ تقرير حالة؛ الإمارات العربية المتحدة.

Trichilemmal (pilar) cysts mainly occur in areas of dense hair follicle concentration, with 90% arising in the scalp and the other 10% occurring on the face, neck, torso and extremities.[1,2] These benign encapsulated lesions are seen in adults and are solitary in only 30% of patients.[1,2] A familial predisposition to these cysts has been recognised with an autosomal dominant pattern of inheritance.[3] Familial trichilemmal cysts occur in patients younger than 45 years old and are usually large (>5 cm), either solitary or multiple and have histological features of proliferation and ossification.[3] Proliferating trichilemmal cysts are progressive slow-growing nodules commonly seen in women with a mean age of 65 years.[4] Trichilemmal cysts have been known to occur in atypical locations with no hair follicles, such as on the pulp of the fingertips.[5–7] The first case of a penile trichilemmal cyst is reported here and the differential diagnosis elucidated.

Case Report

A healthy five-year-old Caucasian boy presented to the Cooper Health Clinic, Dubai, United Arab Emirates, in March 2012. He presented with an asymptomatic progressive slow-growing mass over the ventral aspect of the *frenulum* of the penis. He had undergone a modified tubularised incised urethral plate urethroplasty for subcoronal hypospadias at the age of 18 months [Figure 1].[8] The distal hypospadias repair involved performing a longitudinal incision on an adequate urethral plate. The incisions in the lateral *glans* were made longitudinally, wide enough to allow two strips of epithelium for a size 10-Fr catheter in the neo-urethra. The neo-urethra was tubularised in the midline in two layers and was subsequently reinforced with a dartos flap. A waterproof layer harvested from the hooded *prepuce* and juxtaposed ventrally completed the repair. The *glans* wings were closed in the midline and a meatoplasty ensured a ventral slit in the glanular *neomeatus*. The hooded foreskin was excised and the skin closure was completed.

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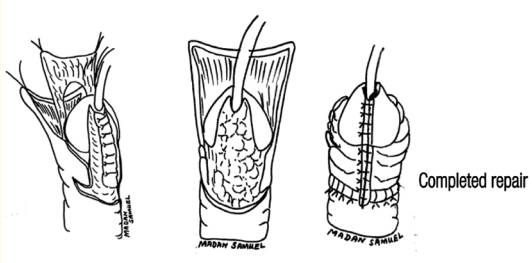

Figure 1: Illustration of a modified incised tubularised urethral plate repair. A vascularised waterproof layer is harvested from the dorsal aspect and buttonholed and transposed ventrally to cover the neo-urethra. Glanuloplasty, meatoplasty and hemi-circumcision are then performed to complete the repair.

Following the repair, the child had a ventral slit in the glanular *meatus* and a circumcised penis. At three-, six- and 10-month postoperative follow-ups, the child was observed to have a functional circumcised penis without complications [Figure 2].

At 30 months of age, the patient had a visible mass which had slowly but progressively increased in size. The mass was soft, cystic, smooth-surfaced, elastic, non-tender and relatively mobile, measuring 1.5 x 1.6 x 1.5 cm. It was not inflamed and no *punctum* was visible. Following a provisional diagnosis of an epidermoid cyst, the mass was excised under general anaesthesia. Excision involved the removal of the intact cyst which extended below the *glans* penis. Proximally, the ventral aspect of the *glans* was incised longitudinally and the two glanular wings were mobilised laterally away from the capsule of the cyst. A hemi-circumferential transverse incision was made distally at the level of the mucosal cuff and the penile shaft skin. The skin was mobilised away from the capsule of the cyst. Following the excision of the intact cyst, an iatrogenic hole was

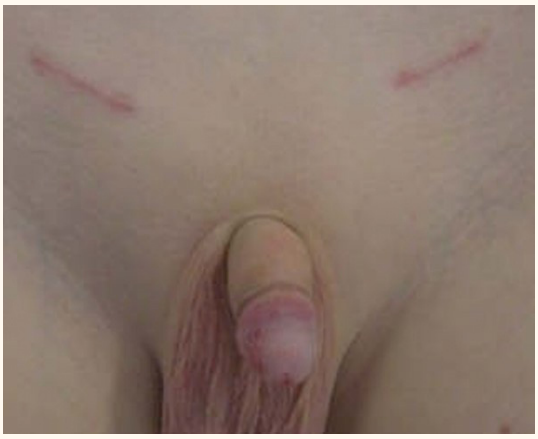

Figure 2: Photograph of the functional circumcised penis post-hypospadias repair. The child also underwent a bilateral inguinal herniotomy. Inguinal hernias can occur in association with hypospadias in 7–13% of children.[8]

noticed in the urethra at the coronal level. Proximally, 2 mm of the *meatus* was excised with the cyst. The dorsal nodular skin was also excised and the hole in the urethra was closed in two layers. Neoglanular *meatus* was created and the glanular wings were approximated in the midline. Skin closure was completed by suturing the skin to the mucosal cuff.

Histology revealed that the cyst was surrounded by a fibrous capsule that was lined by a palisade of small cuboidal basal epithelial cells with no intercellular bridging. No granular cell layers were seen. There were *foci* of calcification, mitoses and keratinisation. There was no cell *atypia*, cellular necrosis or cyst wall penetration. The keratin was stained with anti-keratin antibodies derived from human hair. The histological diagnosis indicated a trichilemmal cyst [Figures 3A & B].

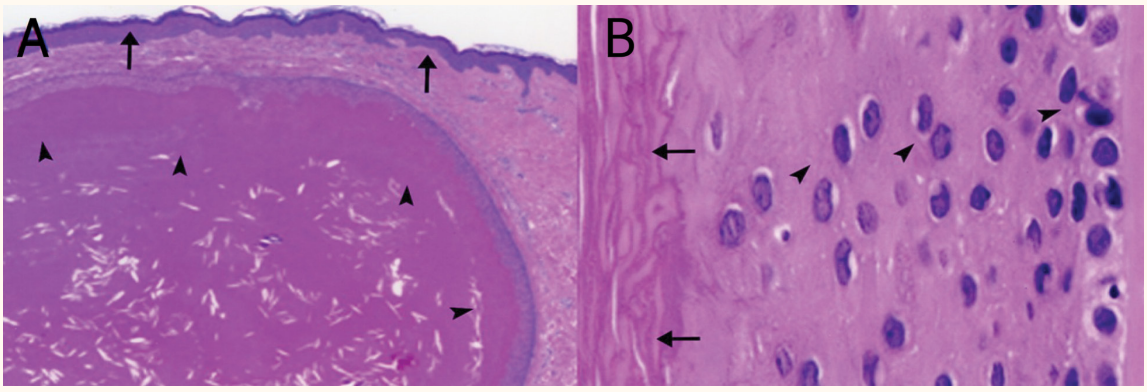

Figure 3A & B: Histology of the encapsulated benign trichilemmal cyst showing the eosinophilic centre lined by walls of stratified squamous epithelium. **A:** Haematoxylin and Eosin (H&E) stained specimen at x100 magnification showing calcification and keratinisation (arrowheads), as seen by the lobulation of the cyst wall with bulwarks of squamous epithelium. The arrows show the palisades of squamous epithelium without a granular layer. **B:** H&E stain x1,000 magnification with oil immersion highlighting the palisades of squamous epithelium (arrows) and keratinisation with mitotic figures (arrowheads). These features are characteristic of a trichilemmal cyst.

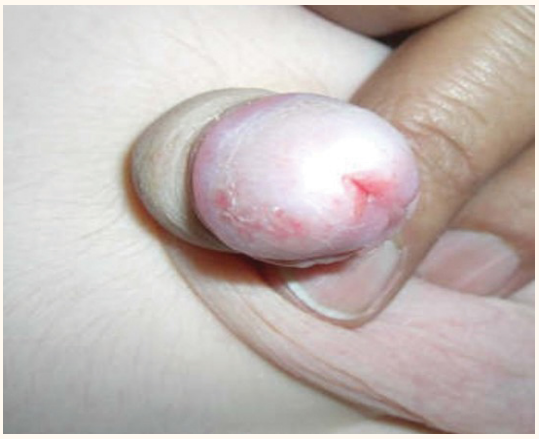

Figure 4: Photograph of the patient's normal circumcised penis with a ventral slit in the glanular *meatus* after the excision of the trichilemmal cyst.

At a 26-month post-excision follow-up, there was no evidence of recurrence or other complications. Figure 4 shows the patient's normal circumcised penis with a ventral slit in the glanular *meatus* after the excision of the trichilemmal cyst. A retrospective in-depth family history revealed the absence of trichilemmal cysts occurring in any other members of the family (either paternal or maternal).

Discussion

This is the first report of a penile trichilemmal cyst occurring in a paediatric patient after a hypospadias repair. Trichilemmal cysts are lined by stratified squamous epithelium. The squamous epithelium is analogous to that seen in the isthmus of the hair follicle. The isthmus bridges the erector *pili* muscle and the sebaceous gland duct.[1–4] An inner root sheath is absent and the squamous epithelium undergoes rapid keratin formation without a granular cell layer. These are the characteristic findings of a benign, non-inflamed, non-infected and non-malignant trichilemmal cyst.[1–4] The contents of the cyst may extrude to form a soft cutaneous horn.[9,10] Although they occur mainly in hairy areas such as the scalp, cysts can arise on the face, neck, extremities and in atypical areas that are devoid of hair follicles, such as the fingertips.[2,5–7] In the current patient, squamous epithelial *metaplasia* with associated keratinisation at the isthmus, likely due to the hypospadias repair, resulted in the formation of a trichilemmal cyst. A complete excision of the cyst, as in this case, prevents recurrence. Moreover, early excision of these cysts is recommended in order to prevent infection, inflammation and, in rare cases, malignant changes.[11]

Penile cysts are usually solitary and occur in various sizes.[1–4] The differential diagnoses of cystic lesions of the male genitalia are varied and wide-ranging. Critically, lesions that can occur after a hypospadias repair include urethral diverticula, ureth-rocutaneous fistulae and, less commonly, epidermoid cysts.[8] Epidermoid cysts can be congenital or acquired; acquired epidermoid cysts occur following mechanical implantation of epidermal fragments.[1–4,9] The latter was a possibility in the current patient due to his previous history of a distal hypospadias repair. Therefore, the initial working clinical diagnosis was of an epidermoid inclusion cyst following the repair. Histopathology provided the definitive and conclusive diagnosis of a trichilemmal cyst. It was hypothesised that the distal hypospadias repair had triggered squamous *metaplasia* with keratinisation, leading to the development of a trichilemmal cyst in a non-hair-bearing area of the body.

Early and complete excision of penile cysts are indicated to prevent the risk of urethral obstruction, infection, inflammation, proliferative growth, impediments to coitus in the future and malignant changes as well as for aesthetic purposes.

Conclusion

This case report adds a rare diagnosis of penile cysts to the literature, as this is the first reported incidence of a penile trichilemmal cyst occurring in a paediatric patient after a hypospadias repair. Early and complete excision is recommended to prevent complications and recurrence.

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