



Hair tourniquet syndrome of the clitoris and labia minora: Report of three cases and review of the literature

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ABSTRACT

Hair tourniquet syndrome (HTS) is a rare condition and may pose diagnostic dilemma. The purpose of this study is to report our experience in 3 female cases with HTS involving genitalia. All the patients in this series presented with swollen and edematous genitalia and clitoris accompanied by strangulation in clitoris or labia minora with a hair thread. Strangulating hair was excised and removed under local anesthesia. The pain was cleared away and edematous discoloration disappeared immediately. Local application of antibiotic ointment was added to the treatment in these cases for a period of 6-7 days. The anatomy of the genitalia was found to be normal in these children during the follow up exam. A brief literature review on this subject is also given in this review.

Key Words: Hair tourniquet syndrome, clitoris, labia minora, strangulation.

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Introduction

Hair tourniquet syndrome (HTS) may be a challenge to the health providers. In this condition, a part of the body is firmly encircled by a various materials in a circular manner. These materials can be hair, thread or similar object. Most sites of involvement in body are fingers, toes and genitals. In order to attain favorable outcome and avoid hazardous

consequences of this rare disease such as autoamputation in affected body part, prompt diagnosis and treatment is vital [1-5]. The purpose of this study is to report our practice in 3 female cases with HTS involving genitalia. The presentation, and management of these patients with a brief literature review on this topic is given in the current study.

Case reports

In this case series study, pediatric patients who were diagnosed and under follow-up for HTS, between June 2017 and May 2019 were included. Along with demographic and clinical characteristics, treatment and follow-up records were collected. In all patients, the

diagnosis of HTS was confirmed by physical examination.

This study was performed in accordance with the 1964 Helsinki Declaration and was approved by the ethical committee of Tepecik Training Hospital, and the written consent from the families of the cases were taken.

Case 1

A previously healthy 8-year-old girl was admitted to our clinic with severe pain and swollen vulva for 3 days. On physical examination she was otherwise normal. In the perineum, labia majora and clitoris was found to be swollen and edematous and hair strangulation at the base of the clitoris was detected (Fig. 1).



Fig. 1. Preoperative view showing the encircling hair at the base of clitoris. (Arrow: encircling hair).

After excision and removal of the strangulating hair, the pain was relieved and edematous discoloration disappeared dramatically. Local treatment with antibiotic ointment was continued for 5 days after excision. On follow-

up exam 7 days later, the anatomy of the genitalia had returned to normal (Fig. 2).



Fig. 2. Postoperative view 3 days after removal of the hair.

Case 2

A 7-year-old girl having pain and swollen vulva for 4 days' duration was admitted to our clinic. Perineal inspection revealed a swollen and edematous labia majora with a hair strangulation at the base of the clitoris. Strangulating hair was excised under local anesthesia and the pain disappeared promptly. After a local treatment with antibiotic ointment for 6 days, the anatomy of the genitalia had returned to normal.

Case 3

A 9-year-old girl with a swollen and painful labia minora was admitted to our department. Physical examination revealed a hair thread strangulating labia minora and it was found that the clitoris was not involved. Strangulating hair was excised and removed under local anesthesia. The pain was cleared away and local application of antibiotic

ointment was added to the treatment for 6 days. The anatomy of the genitalia was found to be normal during the follow up exam on 7th day. With an uneventful postoperative period, the family of the child is happy for their child's genital appearance.

To sum up, our management in the presented study included removal of hair coil from the genitalia of our patients under local anesthesia with topical prilocain using a clamp and scissors. Following this procedure edema resolved suddenly. Local treatment with antibiotic ointment was also found to be helpful in relieving the edema and inflammation after excision of the strangulating hair coil.

Discussion

HTS is an uncommon condition rarely seen in children. In this disease the appendages are strangulated by a hair, a thread, or a fiber in a circumferential manner [1]. Guillimeau first described this entity in 1612. The condition was documented and published in 1832 [2, 3]. Different terminologies for description of this scarce entity exist. These nomenclatures hair tourniquet, coil strangulation, toe tourniquet and hair thread or acquired ring syndrome [4, 5].

As a rule any part of the body may be affected by HTS. Fingers, toes and penis are usually affected [6, 7]. Less commonly, there may be other parts of the body that can be involved by the disease and these sites are nipple, tongue or uvula, clitoris, labia, ear lobes [1, 8-13]. In a study with 210 cases of HTS, the different parts of the body have been affected by the disease in a decreasing order of frequency and of the patients 44.2% had penile HTS, 40.2% had involvement of toes by the disease, 8.6% had HTS of fingers and 6.8% of the patients presented HTS in other sites [1].

Most commonly young children are affected by HTS. The age range of the patients differ with respect to the site of the body involved by the disease. It has been reported that age ranges of patients with finger HTS and penile involvement have been found to be between the neonatal periods of life up to 1.5 years and 4 months to 6 years, respectively [14].

With respect to labial and clitoral involvement by HTS, older age groups of children have been affected more compared to younger offsprings (age 7-13 years) [15]. Despite the limited number of our cases with HTS, the average age of 8 years at presentation in the presented study is in accordance with previous reports [15, 16].

The pathological explanation of HTS lies on the high traction resistance of hair making it as an effective turnstile. Besides moisture of the environment has an additive effect on this strain power [4]. The length of the hair changes depending upon the degree of humidity and hair elongates if there is much humidity while it constrains back to its normal size if there is no humidity in the environment. Thus the dry environment causes hair fiber to strangulate around an appendage causing the affected appendage to be hypoperfused [17]. Firstly constricting hair reduces the venous and lymphatic drainage causing edema. Later on, if no precaution is taken, arterial supply to the affected body part reduces leading to ischemia even autoamputation. It has been reported that this process of autoamputation can occur within hours to weeks [2, 18]. For this reason it is paramount to recognize and manage these cases in order to prevent deficiency of function or decapitation of the affected apart of the body [19, 20].

Differential diagnosis of HTS contain variety of diseases. These are infection, trauma, and bites by insects, dermatitis, keratoderma and

congenital bands [4]. Other disease states that should be differentiated from HTS include exploitation of child, ainhum that is the digital constriction affecting a toe, ainhum like states and paronychia [4, 5]. There are various predisposing factors in HTS and one of them is "teleogen effluvium". Because of maternal hormonal changes most of the mothers, during postpartum period, come up with excessive hair loss and this is called teleogen effluvium [21]. The babies of these mothers with teleogen effluvium are in danger of HTS and typically these children are younger than 4 months [21].

The gold standard in the treatment in HTS is prompt removal of the compressing fiber. There are various techniques of surgical intervention reported in the literature and these are detaching if there is minimal edema, cutting out of the tight stretched hair with scissors or scalpel blade. Depilatory creams are safe choice of treatment and they do not produce any discomfort but their usage is not approved [22]. Especially in late diagnosed patients having HTS surgical debridement is another option [1, 23].

Conclusion

HTS may cause diagnostic uncertainty for the clinicians dealing with these patients. If a child presents with a swollen appendage in any part of the body HTS should also be kept in mind and the medical providers should be cognizant of this rather rare entity. Children with HTS can be prevented from unwanted consequences of this entity and treated if these cases are diagnosed early and managed appropriately. In order to avoid undesirable outcomes like decapitating injuries, it is paramount to diagnose these children promptly and timely treatment is essential.

Compliance with ethical statements

Conflicts of Interest: None.

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Consent: All photos were taken with parental consent.

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