

A Case of Genital Hair Tourniquet Syndrome and Review of the Literature

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Received: July 14, 2017; **Published:** August 17, 2017

Abstract

Hair tourniquet syndrome (HTS) is a rare clinical phenomenon. In this condition, a body appendage is tightly and circumferentially wrapped by hair, thread or similar material. Prompt diagnosis and treatment of this condition is vital to attain good outcome and prevent even a catastrophic consequence of autoamputation in affected body part. We here report a case of HTS involving genitalia in a 8-year-old girl. It is also aimed in this report to review current information about management of HTS in the light of relevant literature.

Keywords: Hair Tourniquet Syndrome; Catastrophic; Toes; Fingers

Introduction

Hair tourniquet syndrome (HTS) is a rare clinical phenomenon. In this condition, a body appendage is tightly and circumferentially wrapped by hair, thread or similar material. Commonly affected sites include fingers, toes and genitals. Prompt diagnosis and treatment of this condition is vital to attain good outcome and prevent even a catastrophic consequence of autoamputation in affected body part. We here report a case of HTS involving genitalia in a 8-year-old girl. It is also aimed in this report to review current information about management of HTS in the light of relevant literature.

Case

A previously healthy 8-year-old girl was admitted to our clinic with a complain of severe pain and a swollen vulva of 3 days' duration. 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 (Figure 1). 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 (Figure 2).



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



Figures 2: Postoperative view 3 days after removal of the hair. The patient is under local treatment with antibiotic ointment.

Discussion

HTS is an uncommon acquired condition where appendages are strangulated by an encircling strand of hair, a thread, or a fiber [1]. First description of a tissue strangulated by a thread of hair was in 1612 by Guillimeau and the first documented report of this condition was published in *Lancet* in 1832 [2,3].

There are a number of pseudonyms used for definition of this clinical entity. These are namely tourniquet syndrome, toe tourniquet syndrome, hair thread tourniquet syndrome, hair tourniquet syndrome, hair coil strangulate syndrome or acquired constriction ring syndrome [4,5]. Essentially, any appendage may be involved by this disease. Commonly affected parts of the body include fingers, toes and penis [6,7]. It has also been reported that other body parts including clitoris, labia, ear lobes, umbilicus, nipple, tongue or uvula may be involved by this disease [1,8-13]. In a meta-analysis comprising 210 cases of HTS, 44.2% involved penis, 40.2% the toes, 8.6% fingers and 6.8% represented other sites [1].

Most cases of HTS occur in young children. Observed age range of reported cases with finger HTS is between first days of life up to 19 months and penile involvement is 4 months to 6 years [14]. Labial and clitoral wrapping have been described in an older age group (age 7-13 years) [15]. Concerning the age at presentation, the presented case in this report with an age of 8 years is similar to those reported previously [15,16].

The high tensile strength of hair makes it an effective tourniquet and humidification of the hair has an effect on the tensile strength [4]. When wet, hair stretches out, and when dry it constricts back to its normal size. Thus if a hair is wrapped around an appendage, it may cause strangulation of the affected appendage when it dries [17]. In pathophysiological point of view, first constricting hair results in reduced venous and lymphatic drainage causing edema. If untreated, raised interstitial pressure may reduce arterial supply, causing ischaemia of the affected body part. Delay in diagnosis and treatment may cause catastrophic consequence of autoamputation of the involved part of the body and this process can occur over hours to weeks [2,18]. Prompt recognition and timely management of these cases is important to prevent loss of function or autoamputation of the involved appendage [19,20].

Differential diagnosis of HTS includes infection, trauma, insect bite, allergic or irritant dermatitis, palmoplantar keratoderma and congenital constriction bands [4]. Child abuse, ainhum (digital annular constriction affecting a toe), pseudoainhum and paronychia should

also be considered in the differential diagnosis [4,5]. As a one of the predisposing factors in HTS, “teleogen effluvium” deserves special attention. During postpartum period, 90% of mothers experience excessive hair loss called teleogen effluvium due to maternal hormonal changes [21]. This situation subsequently exposes their infants to the risk of HTS. It has been reported that the children with HTS due to teleogen effluvium are typically younger than 4 months [21].

Removal of the offending fiber as soon as possible is the corner stone in the treatment of these patients. Reported techniques of surgical intervention include unwrapping method in cases with minimal edema, cutting of encircled hair with scissors or scalpel blade. Although its usage is off-label, use of depilatory creams has been reported to be safe alternative to instrumentation with minimal discomfort [22]. Cases requiring surgical debridement have also been reported for the treatment of late diagnosed patients with HTS [1,23]. Under local anesthesia with topical prilocain, hair coil was easily removed by a clamp and scissors from the genitalia of our patient and edema resolved dramatically after the procedure.

HTS is a rare disorder and it is frequently a diagnostic dilemma for the front liners of medical providers. It should be considered in the differential diagnosis of swollen appendages and the clinicians dealing with these children should be aware of this uncommon entity. HTS is a preventable and treatable condition if diagnosed early and managed appropriately. To avoid constrictive injury such as ischemia and autoamputation of the involved body part, prompt recognition and timely treatment is a necessity rather than of choice and may prevent decapitating injuries.

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Volume 5 Issue 2 August 2017

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