The relationship between the localization and etiology in children’s penile tourniquet syndrome: A case report and literature review

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Abstract
Though the penile tourniquet syndrome is rarely seen, it is a disease which causes complications from a simple edema to severe necrosis and amputation. Whether simple or complicated, the penile strangulation that is the causative factor, except babies, whom are usually questionable. Our aim is to investigate the relation between perpetrators and location of disease in the penile tourniquet syndrome.

Key Words
Penile tourniquet syndrome; child; location; perpetrator.

INTRODUCTION
Penile tourniquet syndrome (PTS) is a rare condition. PTS is featured by progressive penile strangulation led by constriction of the penile shaft [1]. It causes complications from a simple edema to urethrocutaneous fistula or severe necrosis and amputation [2,3]. PTS was caused by sexual fantasies in adults and mother's hair in babies. However, in cases between these two age groups, both patient and his family do not give convincing information about the etiology [2-4]. Although diagnosis and treatment of PTS have been described by the authors in the literature, the relationship between
perpetrators and location of PTS haven’t been analyzed enough till now. In this study, we sought to explain the relationship between them.

**CASE REPORT**

An 8 year old boy presented with pain, swelling and red discoloration of his penis for three days associated with intermittent difficult micturition and abdominal pain. Even though he was a shy and stutter child he admitted that he tied the radix of penis three days ago when he was playing “knotting game” and told his mother three days after. Fortunately, the ring of tightly tied thread at the radix of penis had been promptly unwrapped by his mother. On examination, there were narrowed urethral meatus, a circumferential 2-3 millimeters epidermal laceration at root of his penis. There was no urethral injury at all, but edema and red-purple discoloration of the whole penis (Fig.1). This infected wound was locally treated with debridement and dressing with povidine iodine. The patient comfortably started to urinate after single dose of intravenous prednisolone. After then, he was treated with oral ibuprofen and ampicillin, and discharged. Three weeks later, the patient almost recovered (Fig.2).

**DISCUSSION**

In the review of literature, between the years of 1967-2014, we saw that the penile strangulation in babies was especially caused by mother's hair which is called telogen effluvium. The hair loss commonly
experienced by women after giving birth, has been known to predispose the risk of PTS. It often happens between 2 and 6 months postpartum [1,4]. It was first reported in 1883 and the other reasons of PTS were introduced in the second half of twenty-first century. The most frequent agents responsible for PTS in the other ages of children (especially ranging from 4-11 years old), which corresponds to the Sigmund Freud’s phallic stage of psychosexual development, are rarely hair, but often fabric threads [1-3]. In contrast to children in adults, the agents are generally metallic (rings etc.) or nonmetallic (condom ring etc.) which placed around the penis to magnify sexual intention [2]. The duration of penile tourniquet may be short (from ranging hours-days) or long (1-7 years) [2,6]. As the duration of PTS prolonged, complications varies from edema to urethrocutaneous fistula or severe necrosis and amputation. Injuries associated with PTS are rare but can cause critical outcome. Hence, it should be treated as soon as possible. The management of PTS contains two stages: removal of the object causing strangulation and treatment of the complications (e.g. urethrocutaneous fistula, gangrene or amputation) because of strangulation [2,3].

The location of the strangulation in our patient was in penile radix, and to our knowledge, it was the first case in the literature. When we reviewed the reported PTS cases, the strangulation was located in the coronal sulcus in 37 cases [3, 7, 11-13]. In the 36 cases, the location wasn’t clear, it would be in the midpenile localization or coronal strangulation by all accounts or according to figures of papers [2]. The location wasn’t defined in the 4 cases but there were suspects of child abuse and neglect that couldn’t be proved by physicians [4, 8]. In 3 cases of midpenile strangulation, the first case had admitted that he tied his penis due to enuresis, the second had admitted, he tied but there was no reason, and third had not admitted anything at all [1,6,9]. In our case, although he was a
shy and stutter child, he admitted that he tied the radix of penis when he was playing “knotting game”.

The reports in the literature frequently mentioned the diagnosis and treatment of PTS, but it wasn’t sufficiently stressed and investigated what was the perpetrator and if there was an intention of malevolence or not in the etiology [1-13]. The coronal sulcus and the proximal region were the localizations to be tied easily in many PTS cases, which were strangulated by hair (telogen effluvium) and the cases with suspicion of enuresis or urinary incontinence [3, 4, 7, 8, 10-13]. The localization of PTS between the penile radix and proximal penis revealed admittance or suspicion of self-inflicted strangulation [1,2,6,9].

With the help of this knowledge, we postulate that if the localization of PTS is the coronal sulcus and distal part of it, the perpetrator may be either the hair of mother (telogen effluvium) which is accidental or the deliberate cases because of enuresis or urinary incontinence. If we exclude the accidental cases, when the intention was punishing, someone may have strangulated the coronal sulcus and distal part of the penis. On the contrary, if the localization of PTS is between the penile radix and mid-proximal penis, the perpetrator may have been the patient himself as it was happened in our patient. Because penile amputation can be the result in PTS, those clues may play an important role in the forensic medicine.

CONFLICT OF INTEREST
None declared.

REFERENCES


