Hair-thread Tourniquet Syndrome causing uvular strangulation in a child

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DESCRIPTION

A normally fit and well 3-year-old girl awoke distressed. Her parents identified a long hair protruding from her mouth. She was comforted and given some food and drink. Afterwards they could no longer see the hair and she had no ongoing pain or issues initially. Over the subsequent days, she had increasing difficulty swallowing solid foods due to pain on swallowing and the parents later noticed a discoloration of the tip of her uvula. The family attended a walk in centre, the general practitioners and subsequently the children’s emergency department 3 days after the initial incident. She was reviewed by the ear, nose and throat team who found that the distal tip of her uvula was white and that a hair could be seen trailing from it inferiorly. History and examination were otherwise unremarkable.

The child remained well and was taken to theatre for examination under general anaesthetic. On examination in theatre, the hair was found to be strangulating the distal uvula and trailing into the oesophagus (figure 1). It was not possible to untangle the hair, and as the tip was non-viable, the decision was taken to remove the uvula tip with bipolar cautery to reduce discomfort (figure 2).

The child was well postoperatively and discharged home the same day. Telephone follow-up with the parents at approximately 1 month post procedure confirmed no ongoing symptoms or complications.

Hair tourniquet syndrome (HTS) is a rare condition, more commonly seen affecting infants’ external appendages, such as digits and genitalia. 1 Oral HTS has been described rarely, affecting sites,

Patient’s perspective

My daughter uncharacteristically awoke one morning crying. Unable to comfort her, I identified a long hair protruding from her mouth, which caused her severe distress if touched. Unable to adequately examine her mouth, I assumed the hair was caught around a tooth. Fluids and a banana soothed her distress and allowed me to check her mouth where I could no longer see any sign of a hair. Over the following 2 days, she was well but mentioned discomfort when eating. I regularly tried to examine her throat but was not confident that the hair was wrapped around her uvula, partly as it was difficult to examine and her hair is blonde but also due to my assumption that such an event was highly improbable. However, when I started to witness a discolouration in the tip of her uvula, I sought medical assistance. Although the uvula was discoloured, a minor injuries unit dismissed the likelihood of Hair Tourniquet Syndrome, but following this up with our general practitioner led to a hospital admission and surgery. Following discharge, my daughter had no ill effects in either the short term or long term, awaking the following day without the need for pain relief or a change in diet. I assume her naturally long curly hair attributed to this incident.

Learning points

► Hair tourniquet syndrome (HTS) is a rare cause of strangulation of appendages.
► Swallowing difficulty with a history of a hair in the mouth should prompt consideration of oral HTS.
► Management of oral HTS will depend on the appendage involved and viability of the strangulated tissue. Under anaesthesia, options include either removal of the hair to allow for tissue reperfusion or autoamputation, or surgical debridement of the tissue if it is clearly non-viable.
Images in... including the teeth, tongue and uvula. A systematic literature review demonstrated only three published cases of uvular HTS in the past 40 years, all in infants. In these previously documented cases, the hair was removed and the uvula allowed to reperfuse or autoamputate. Manipulating a knotted hair in the oral cavity is challenging, and there are no documented cases of the hair being removed without sedation/general anaesthesia.

The uvula is a posterior extension of the soft palate which prevents nasal regurgitation on swallowing and resonates during speech. Uvular necrosis is most commonly iatrogenic, following intubation, endoscopy or suction. Most documented cases, predominantly in adults, resolve conservatively, with necrotic tissue sloughing off in 7–14 days.

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