

A case report with sacral appendage: Is it accessory penis or human tail?

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Received: 16th December 2016, Revised: 9th January 2017, Accepted: 10th January 2017

SUMMARY: Kocaoğlu C, Kocaoğlu Ç, Akkoyun İ, Karaniş İE. A case report with sacral appendage: Is it accessory penis or human tail? Turk J Pediatr 2017; 59: 335-337.

Lumbo-sacro-coccygeal appendages are very rare congenital anomalies. It is difficult to say that they are a evolutionary inheritance (tail) from our ancestors or an anomaly (pseudotail) occurring during embryonal development. If it is a true tail, this lesion contains adipose and connective tissue, striated muscle, blood vessel and nerves, and is covered by skin. However, if this lesion is a pseudotail, it may be an anomalous prolongation of the coccygeal vertebra, lipoma, teratoma, condrodystrophy or parasitic fetus.

We present an infant with a sacral appendage resembling a penis, and its clinical and pathologic differential diagnosis and management are discussed based on literature. Sacral appendages, such as accessory penis or human tail, are treated through simple surgical excision. However, patients must be carefully evaluated regarding teratoma and spinal cord pathology.

Key words: accessory penis, human tail, infant, sacral appendage.

Lumbo-sacro-coccygeal appendages are very rare congenital anomalies. These anomalies are described as a remnant of a structure found in embryonic life or ancestral forms and can be related with other pathologies of the spine and spinal cord, such as spinal dysraphism, meningomyelocele and tethered spinal cord¹. In addition, the appendages may also be associated with an anomalously prolonged coccygeal vertebra, lipoma, teratoma, condrodystrophy or parasitic fetus².

In the present report, we present an infant with a sacral appendage resembling a penis, and its clinical and pathologic differential diagnosis and management are discussed based on literature.

Case Report

Born after an uneventful full-term pregnancy, a 6-month-old male infant with birth weight of 3300 g was admitted to the department with a “finger-like” appendage existing since birth. Physical examination revealed a penis- or tail-like structure on the midline sacral region, which was about 30 mm long, 10 mm in diameter and purple in color at

distal end (Fig. 1). In addition, a spectrum of slight hypertrichosis was present on lumbar region. Considering his age, neurological findings, including anal sphincter tone and developmental milestones were within the normal limits. On external examination, the infant was seen to have normal male genitalia. No cases of similar anomalies were noted in the family members.

As well as lumbo-sacral spine within normal limits on radiography, no occult spinal dysraphism or meningomyelocele was observed on magnetic resonance imaging (MRI) (Fig. 2). Pelvic and abdominal ultrasonography revealed no sign of the extension of the mass or the abnormal formation.

A midline vertical incision was performed by encircling the base of appendage, and the appendage was simply excised. Histological examination of resected specimen revealed a glans-like structure, including nerve, vessel and muscle tissue. No urethra- or testicle-like structure was present (Fig. 3). The infant was discharged on the 3rd postoperative day. During the follow-up period of 6 months, the infant



Fig. 1. Photograph showing the sacral appendages

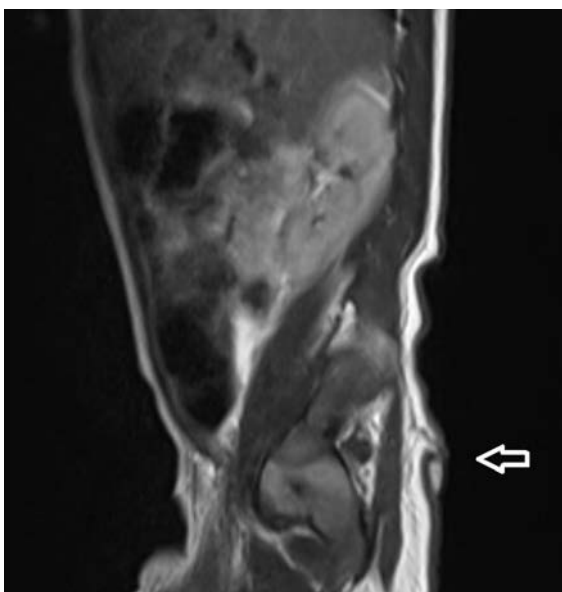


Fig. 2. Preoperative sagittal T1-weighted MR image demonstrating high signal intensity of the tail appendage (white arrow)

displayed normal neurological development and commenced to walk and speak at the 12th month.

Written informed consent was obtained from the patient's mother for publication of this case report and accompanying images.

Discussion

The fetal tails usually disappear by the end of the gestational 8th week in human beings. After delivery, the caudal appendage resembling a tail is a quite rare entity¹. It is difficult to say that the caudal appendage is an evolutionary inheritance (tail) from our ancestors or an anomaly (pseudotail) occurring

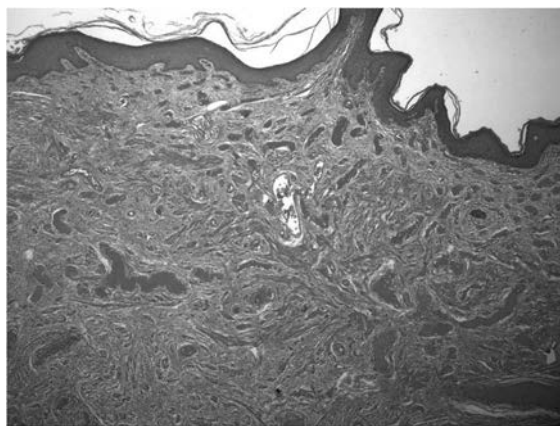


Fig. 3. A polypoid lesion covered by keratinized stratified squamous epithelium and numerous dilated veins, nerves and muscle fibers seen under the epithelium. HEEx100

during embryonal development. If it is a true tail, this lesion contains adipose and connective tissue, striated muscle, blood vessel and nerves, and is covered by skin. However, if this lesion is a pseudotail, it may be an anomalous prolongation of the coccygeal vertebra, lipoma, teratoma, chondrodystrophy or parasitic fetus³.

Teratomas are most commonly seen in sacrococcygium, mediastinum, central nervous system, retroperitoneum and neck, and may also contain a number of diverse tissues and have abnormal tissue relationships and mixtures⁴. According to the findings of histological examination, the tip of the resected specimen obtained in our case consisted of glans-like structure including nerve, blood vessel and muscles. Despite the presence of the findings mimicking most features of a teratoma, the differentiation into glans-like structure did not precisely fit the characteristics of a teratoma. These histologic findings suggested the presence of a penile structure, rather than a teratoma.

Despite histopathological findings suggesting the accessory penis, the morphogenesis of our case was unclear. It can be speculated, however, that during embryogenesis, multipotential cells may mature along the lines of an accessory penis tissue. On the other hand, the sacral appendages may be an evolutionary inheritance, namely a tail, from our ancestors. The localization of the appendage in sacral region and the lack of its connection with vertebrae or neural tissues supported this hypothesis. So, it was difficult to decide whether the case was an accessory penis or human tail.

While the number of cases reporting accompanying additional anomalies is limited in literature, the number of cases including sacral appendage but not accompanied by additional anomalies is even rarer. Upon searching the pubmed by entering the terms “human tails and pseudotails”, we found only two cases published by Wilkinson and Boylan⁵. The human tail or caudal appendage is usually associated with occult spinal dysraphism¹. Spina bifida is the most frequent co-existing anomaly in those with both true and pseudotail⁶. Especially in the cases including the components of neural tissue such as meningocele, surgeons should be alert in order to avoid the sequelae, such as neurologic deficits of the lower extremities, bowel and bladder. Thus, the development of irreversible neurologic deficits may be prevented⁷. In our case, no occult or clear spinal dysraphism, or meningocele was detected on MRI. We considered that the level of anomaly was in the sacral region was effective in the absence of neurologic abnormalities.

In the literature, anomalies coexisting with sacral appendages such as congenital dermal sinus, hydrocephalus, corpus callosum abnormalities, heterotopic gray matter, Arnold-Chiari malformation and tethered spinal cord were reported⁸. In a report of 200 cases by Tavafoghi et al.⁹, the spinal dysraphism was shown to be a condition associated with cutaneous signs in more than 50% of instances. In our case, no other accompanying anomalies except for slight hypertrichosis drew attention on lumbar region.

In such cases, the most appropriate timing for the operation is when the infant is at least 3 months old and/or 5 kg in weight¹⁰. The most appropriate timing for the resection of a human tail is considered to depend on the histopathological structure of appendages and other accompanying anomalies. The case was admitted to our clinic at the age of six months and operated on immediately. No postoperative complications developed.

The treatment of caudal appendages such as accessory penis or human tail not accompanied by other anomalies is simple surgical excision. However, such patients should be investigated carefully in terms of spinal cord pathologies and other accompanying anomalies before the operation.

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