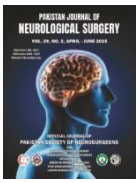

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Case Report

Caudal Appendage in Neonate: Case Report and Literature Review

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ABSTRACT

The caudal appendage, also called a human tail, is a congenital anomaly located in the lumbosacral region, characterized as a prominent lesion. It is one of the lesions considered a marker of spinal dysraphism and may be classified into 2 groups: "pseudo-tail" and "true tail". The authors report a case of a newborn baby girl admitted with the complaint of having a 4 cm long tail that had been present since birth. She was referred to us on the eighth day. The clinical examination found a soft, elongated, skin-covered, and non-fluctuating appendage in the lumbosacral region. She had no neurological impairment. A computed tomography scan showed a lumbar appendage associated with a spina bifida. The tail was surgically removed, and both the surgery and postoperative period were uneventful. Histopathological examination of the tail revealed a benign lesion with mature adipose tissue covered by skin. The caudal appendage is a benign congenital anomaly that may be associated with other spinal cord anomalies. Therefore, neurological examination as well as radiological evaluation is recommended to highlight the underlying anomalies.

Keywords: Caudal appendage, congenital anomaly, spinal dysraphism, human tail, spina bifida.

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INTRODUCTION

The caudal appendage or human tail is a rare congenital abnormality observed in the lumbosacral or sacrococcygeal region. It is a result of abnormal embryological development characterized by a prominent lesion containing adipose tissue, central bundles of striated muscle, and nerves.[1] The first case was documented in 1881. Since that time, cases have been reported sporadically in the literature. This congenital anomaly is one of the lesions considered a marker of spinal dysraphism.[2-4] The authors report a rare

case of caudal appendage associated with spina bifida in a newborn.

Case Report

A newborn baby girl was referred to us with the complaint of having a long tail that had been present since birth. Her mother was a 27-year-old woman who had good antenatal care follow-up, and the pregnancy was uneventful without any comorbidities. Ultrasonography performed during the pregnancy was normal. There was no history of congenital anomalies in the family. The baby was born after uneventful vaginal delivery. On clinical examination, she was alert. She had no fever, and the head circumference was normal. Primitive reflexes were also present. Examination revealed a 4 cm elongated soft, skin-covered, and non-fluctuating appendage. The tail was located in the midline of the lumbar region (Figure 1). There was no voluntary movement of the tail. The child had no neurologic deficit, and both urinary and bowel functions were preserved. A computed tomography (CT) scan showed a lumbosacral pedunculated appendage without bony or cartilaginous elements associated with spina bifida (Figure 2). Under general anesthesia, the baby was placed in the prone position, and surgical excision of the tail was done on day 8. The tail was removed completely after a circumferential dissection (Figure 3). The exploration did not find any nerve or bony element in the tail, and the underlayer fascia was intact. No connection of the tail with the spinal cord was found. The wound was closed in layers (figure 4). Histopathological examination of the tail revealed a benign lesion with mature adipose tissue covered by skin. The postoperative course was uneventful, and the wound healed well with a good cosmetic outcome (Figure 4). Neurological examination was normal.

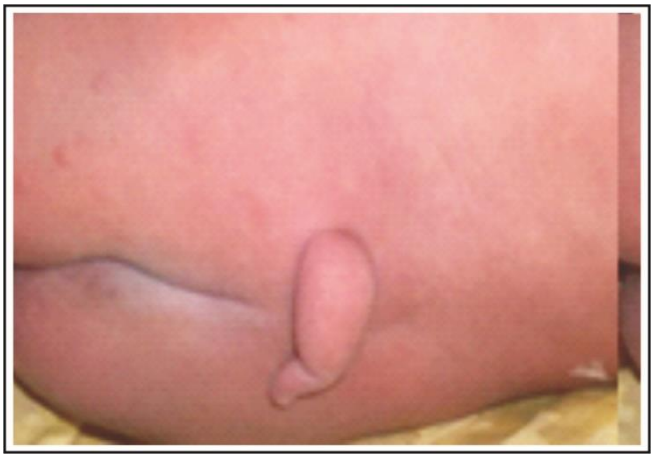

Figure 1: *Preoperative view of caudal appendage located in the lumbosacral region.*

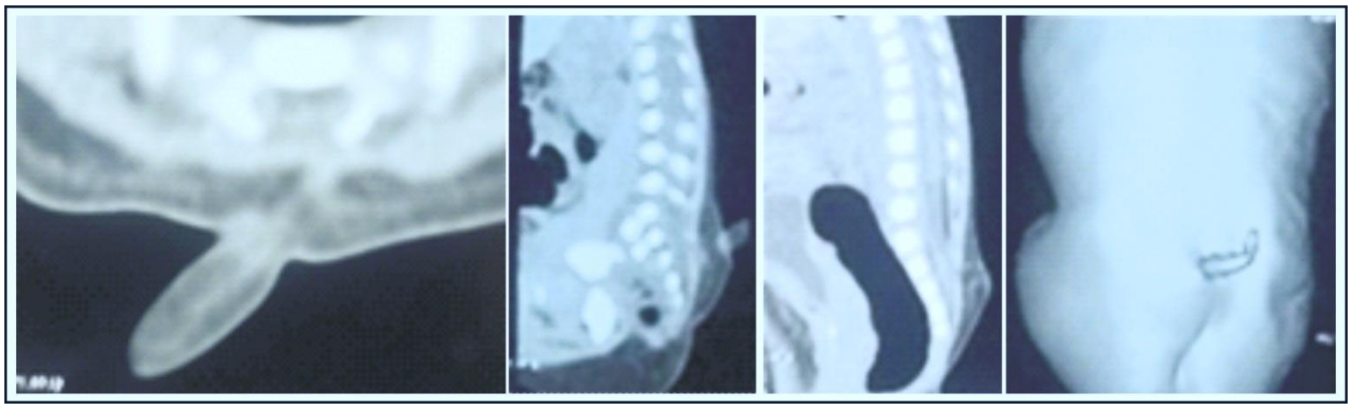

Figure 2: *CT scan imaging showing a lumbosacral appendage without bony or cartilaginous element associated with spina bifida in Axial view (A), sagittal view (B, C), and 3-dimensional view (D).*

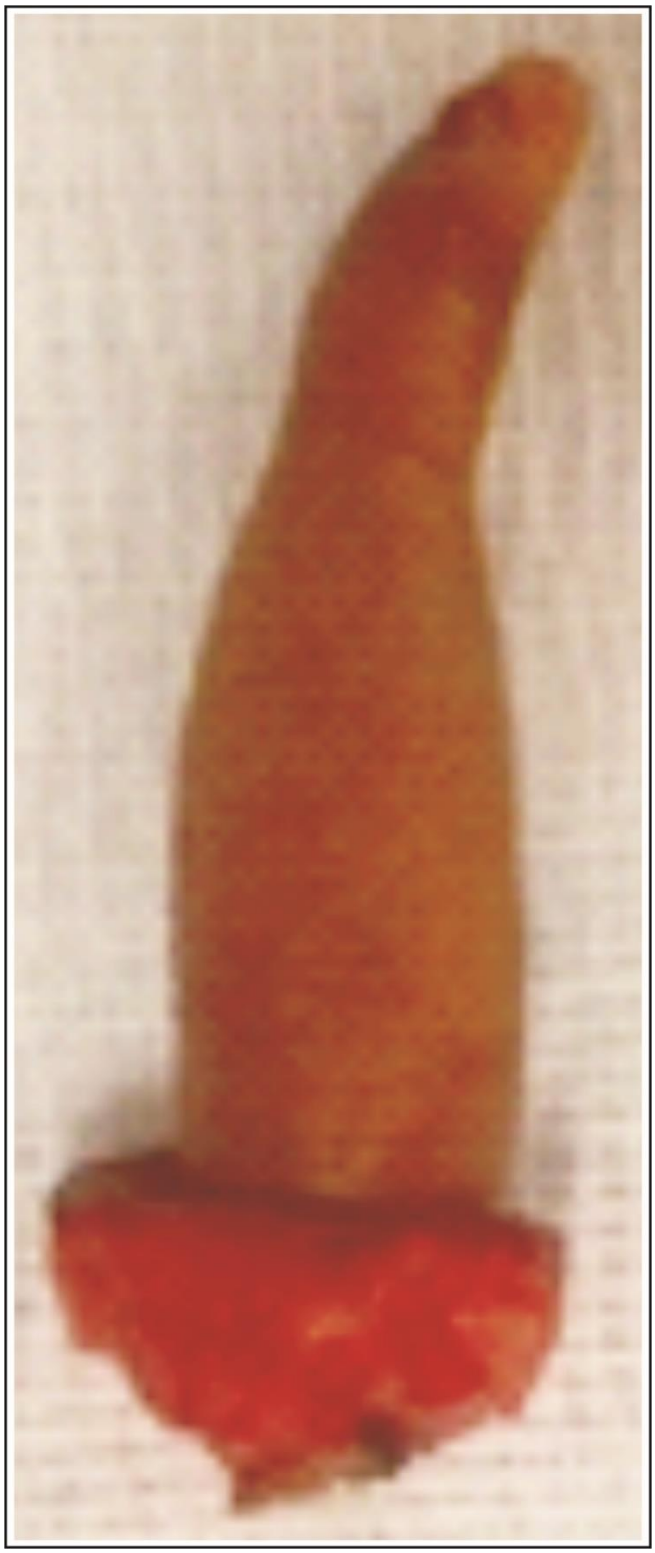

Figure 3: *Photograph showing the tail after surgical excision.*

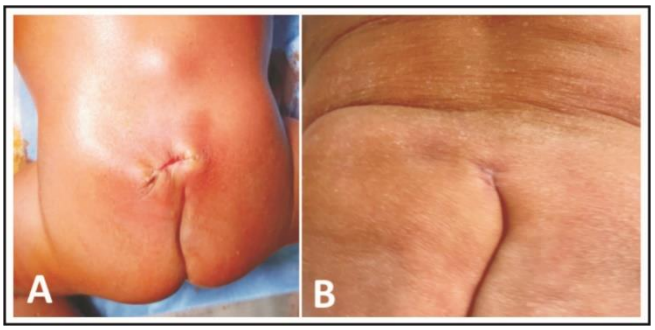

Figure 4: *Photograph showing the wound (A) just after tail excision and (B) 1 month later.*

DISCUSSION

The caudal appendage is a rare congenital abnormality due to abnormal embryological development. It is observed in the lumbosacral region.[3,5] The first case was documented in 1881. In 1984, Dao et al,[6] established a classification into 2 groups, namely: a "true tail" or a "pseudo-tail.[7] A true human tail results from incomplete regression of the most distal end of the normal embryonic tail.[8] Pseudo-tails have no embryological origin[1]. The caudal appendage is found frequently in boys, but our case was an 8-day-old baby girl. Turk et al,[9] have diagnosed and managed five cases of the human tail, and three were male. Although rare, caudal appendage is considered to be a marker of occult spinal dysraphism, in the same way as a tuft of hair or a lumbar hemangioma.[3,10] The presence of one of these elements indicates in 80% of cases the presence of an associated spinal anomaly that should be detected, like spina bifida, split cord malformation, intraspinal lipoma, spinal cord tethering, or myelomeningocele.[11-13] Spina bifida is the most frequent coexisting anomaly.[6,14] Islam et al,[15] found one patient with neurological impairment and spinal dysraphism among four patients with the human tail. In our case, the caudal appendage was associated with spina bifida in CT scan imaging. In a review of 59 caudal appendage cases, Lu et al,[16] found that 81% had spinal tethering in CT or MRI imaging. In our case,

a CT scan did not reveal spinal cord tethering. MRI is the best imaging modality that can highlight these spinal anomalies, particularly the tethered spinal cords. MRI could not be done in our case because it was not available. Therefore, strict clinical monitoring as well as an MRI is recommended to rule out or confirm these anomalies. The caudal appendage can be associated with different rare genetic syndromes, such as Frank Ter Haar syndrome (which combines skeletal, ocular, and cardiac anomalies), Malpuech syndrome (autosomal recessive, with cleft lip, facial dysmorphia), Kabuki syndrome (with facial dysmorphia, skeletal abnormalities and intellectual deficit) or Golz syndrome (focal dermal hypoplasia).[5] Koene et al,[17] reported a case of 3 family generations with a caudal appendage associated with multiple skeletal anomalies due to a pathogenic TAB2 gene. Prenatal diagnosis of caudal appendage has been reported by authors.[18,19] In our case, the clinical examination did not find any other congenital anomalies or genetic syndromes; therefore, genetic testing was not necessary. The aim of surgical management of the caudal appendage is both aesthetic and neurological. Caudal appendage is a benign congenital abnormality.[20] Therefore, caudal appendage without associated spinal cord abnormalities should be managed for aesthetic reasons.[21] Underlying spinal cord anomalies associated with caudal appendage should be managed for neurological considerations, as well as the tail for cosmetic reasons.

Our study is limited by the unavailability of MRI, which is the gold standard for spinal dysraphism. It is recommended in follow-up.

CONCLUSION

Caudal appendage is a benign congenital anomaly that may be associated with other spinal cord anomalies. Therefore, neurological examination as well as radiological evaluation is recommended to highlight the underlying anomalies.

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There is no conflict of interest.

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Additional Information

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AUTHORS CONTRIBUTIONS

Sr.#	Author's Full Name	Intellectual Contribution to Paper in Terms of:
1.	Youssouf Sogoba	1. Study design and methodology.
2.	Jean Marie Kisito Quenum	2. Paper writing.
3.	Seybou Hassane Diallo	3. Data collection and calculations.
4.	Julienne Carole Mouani Ngounou	4. Data collection and calculations.
5.	Boubacar Sogoba	5. Analysis of data and interpretation of results.
6.	Moussa Diallo	6. Analysis of data and interpretation of results.
7.	John N Jabang	7. Literature review and referencing.
8.	Drissa Kanikomo	8. Editing and quality insurer.