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Caudal Appendage: About 3 Cases Report

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ABSTRACT

Introduction: *A caudal appendix is a rare congenital malformation of the median or paramedian diverticulum type located mainly in the lumbosacral region, of soft consistency, exceptionally provided with an axial skeleton and covered with skin often of normal appearance and simulating a tail.*

Patients: *We report the cases of three female patients including two infants aged 4 months and 12 months and an adult aged 35 years. The infants presented caudal appendages located at the atypical breech surface associated with a right cephalocele and the adults had a caudal appendage 11 cm long located at the lumbosacral level 2 cm from the midline on the right. Clinical examination noted one lesion in the first infant and three caudal appendages in the second. The neurological examination was normal. The adult patient did not present any associated clinical malformation. The CT scan noted cranial dysraphism in the form of intra-orbital meningocele associated with shizencephaly for both infants.*

Conclusion: *Caudal appendages are rare and represent markers of dysraphism, which can be cranial or caudal. Their presence requires systematic research through medical imaging examinations.*

Keywords

Caudal Appendage, Congenital Abnormality, Pediatric Surgery, Spinal Anomalies, Neonatal Presentation.

Introduction

A caudal appendix is a malformation such as a median or paramedian diverticulum located mainly in the lumbosacral region, of soft consistency, which may exceptionally be provided with an axial skeleton, covered by skin that is most often normal in appearance and simulating a tail [1]. It is often considered a marker of spinal dysraphia [2-6]. Few extra-spinal cases have been reported in modern literature [4]. We report three new cases including 2 first cases of craniofacial location.

Case Series

Patient 1: A 35-year-old female patient with no known pathological history was seen for a tail-shaped growth implanted in the lumbosacral region that had been present since birth. This malformation increased in volume and size, impacting her social life, forcing her to consult a surgeon before being referred to neurosurgery.

The patient was in good general condition. His locoregional examination noted a pedunculated horn-shaped tumor lesion located in the lumbosacral region opposite the 5th lumbar vertebra and the 1st sacral vertebra (L5-S1) implanted 2 cm from the axis of the spiny on the right. It was cylindrical with a 3 cm wide implantation base and measured 11 cm long. Its consistency was firm, rough and homogeneous with a cartilaginous appearance. There were no active movements. It was entirely covered with epidermises, peeling skin with a “scaly” appearance (Figure 1). The neurological examination was normal. She had no associated malformation. The diagnosis of lumbosacral caudal appendix was made.

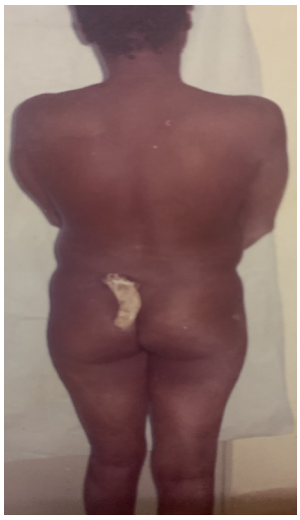


Figure 1: Lumbar caudal appendage.

Paraclinical assessment consisting of a cranioencephalic and spinal scan with and without injection of contrast product as well as a cardiac and abdominopelvic ultrasound were requested to look for associated malformations.
Because she feared and didn't have money, she gave up the hospital and were lost to follow-up.

Patient 2: Female infant aged 12 months was seen for exophthalmos congenital right associated with 3 congenital cheek growths in the shape of a little finger. The evolution was marked by a progressive increase in the volume of the fronto -orbital swelling and the size of the cheek growths.

The child had a good general impression. There was an asymmetry of the cranium more marked on the right with a polymalformative syndrome oculo -facial made of a congenital protrusion of a soft, reddish right intraorbital mass, not very painful and non-hemorrhagic to the touch with an absence of an eyeball and three small congenital pedunculated nodules in the right cheek, the largest of which measured approximately 1cm in diameter and long, of homogeneous soft consistency without active movement; their implantation base was wider and their top rounded. The middle one was the largest (Figure 2). The neurological examination did not note any delay in psychomotor acquisitions or motor deficit.

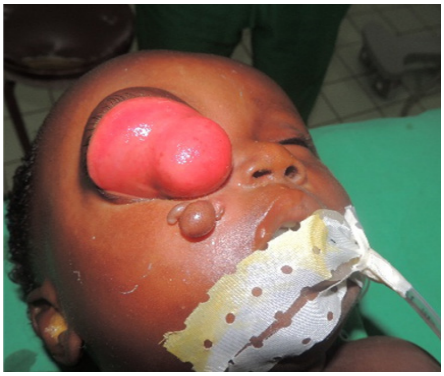
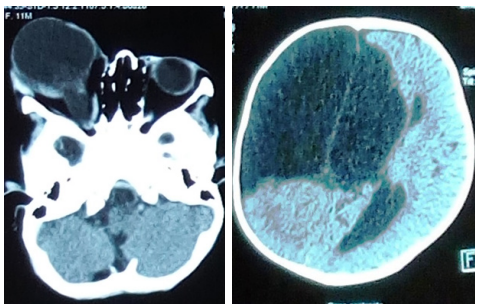


Figure 2: Orbital cephalocele + caudal appendices.

The cranioencephalic scan with and without injection of contrast material revealed a right intraorbital meningocele associated with schizencephaly and a right fronto-temporo-parietal arachnoid cyst. Appendices appeared isolated from meningocele (Figure 3). The paraclinical assessment carried out to look for associated malformations (cardiac ultrasound, abdominopelvic ultrasound, pan-spinal scan) came back normal. The diagnosis of caudal appendages associated with an intraorbital meningocele, a fronto -temporo-parietal arachnoid cyst and schizencephaly was made. The patient benefited from the excision of the caudal appendages and the treatment of the meningocele with simple operative consequences.



a b

Figure 3: Cranio-encéphalic Ct scan.
a: right orbita meningocele; b: arachnoid cyst + schyzencéphaly.

Patient 3: A 4-month-old infant with no particular history of female gender were referred for congenital right orbital protrusion associated with a tail-shaped growth at the sheer. The evolution was marked by an increase in the volume of the protrusion and the tail-shaped protuberance which also increased in size before the date of the intervention due to its young age.

The child had a good general impression. His locoregional examination noted a polymalformative syndrome oculo-facial made of a congenital right orbital swelling associated with a congenital protrusion of a soft and reddish right intraorbital mass not very painful with absence of eyeball and a lateral protuberance in the shape of a tail strongly suggestive of a caudal appendage. The latter was unique with a wide base and a tapered top. This appendix was soft, painless and covered with homogeneous healthy skin without crawling movements. It measured 3 cm long by 1 cm in diameter (Figure 4).

Figure 4: Orbital cephalocele +caudal appendice.

The remainder of the neurological examination was comparable to that of a child his age. The cranioencephalic scan with and without injection of contrast material revealed a right intra-orbital meningocele and shizencephaly associated with a caudal soft tissue appendage (Figure 5).

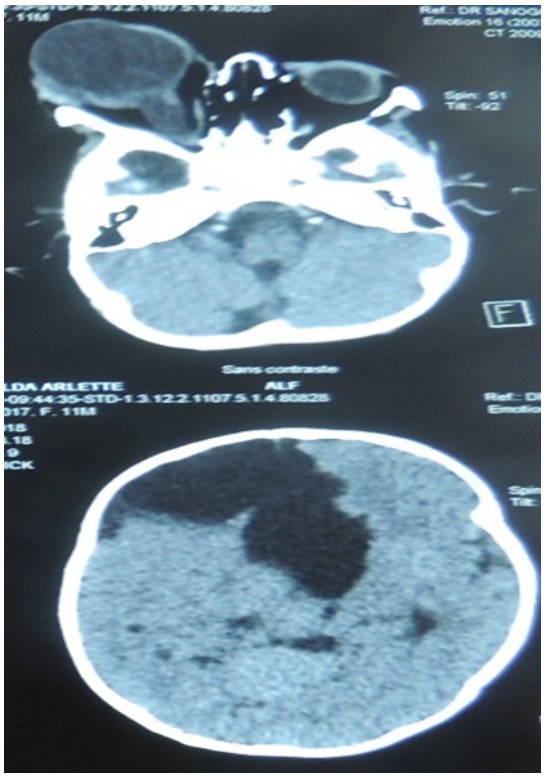


Figure 5: Cranio-encéphalic Ct scann
orbital meningocele (a) + schizencéphaly(b)

The paraclinical assessment carried out to look for associated malformations (cardiac ultrasound, abdominopelvic ultrasound, pan-spinal scan) came back normal. Diagnosis of latero-orbital caudal appendix associated with an intra-orbital meningocele and shizencephaly was made. The infant benefited from an excision of the caudal appendix and a cure for the meningocele with simple operative consequences.

Discussion

The human tail has been mentioned in literature since the early 1900s. These were usually isolated cases that sparked a lot of intrigue, until the end of the 19th century to have observations of a scientific nature [7]. The caudal appendix or "human tail" is a rare anatomical phenomenon whose true incidence remains unknown in the literature due to small published case series. It affects both sexes without any real predominance [8,9] our three cases were all female. All of them had therapeutic itineraries punctuated by orientation errors probably due to the lack of knowledge of the caudal appendages by health workers and the absence of neurological manifestations [3].

The classic location of the malformation is lumbar as in our adult subject. The caudal appendages are generally associated with anomalies including spinal dysraphism [10] and are considered by many authors as cutaneous markers of spinal dysraphism requiring systematic research [6]. Certain atypical locations have been described in the literature at cervical and thigh levels [4]. This motivated Gaskill to prefer the term neuroectodermal appendage because of the close connection between the neuroectoderm and the epithelial ectoderm of the primitive neural tube [1]. Our two infants presented an atypical location of their appendix located at the cranio-facial level. In addition, these appendages were associated with congenital intra-orbital meningoceles representing a rare associated cranial dysraphism [11]. Our two observations represent, to our knowledge, the first cases of cranio-facial localization reported.

Our first patient, 35 years old, presented an increase in size and volume of her caudal appendage during her life which measured 11 cm. Appendages have been described in the literature with variable sizes up to 20 cm [5,12]. The malformation did not present spontaneous movements in our case but it had an atypical skin appearance due to the microtrauma caused by the sitting position.

Many classifications have been proposed since Bartels in 1884, notably that of Dao and Nestky, the best knowned, which divided the caudal appendages into true tails and pseudo-tails. The true tails being remnants of the embryonic tail and the pseudo-tails representing any outgrowth in the lumbosacral region [12]. Our adult patient had a human tail and the infants had pseudo-tails or tail-like structures. All his classifications were of value only for embryology and not for clinical practice [5]. Our two infants presented appendages associated with intra-orbital meningoceles. These appendages could be considered as markers of cranial dysraphism. Many authors have put forward the hypothesis of carrying out a complete assessment in search of malformations located along the cranio-spinal axis. Appendages have been described associated with cranial malformations (agenesis of the corpus callosum, hydrocephalus) [8]. The CT imaging performed revealed in our two infants an intra-orbital meningocele, a shizencephaly without any abnormality located on the spinal axis and an arachnoid cyst in the first child. Imaging is important for the diagnosis of associated lesions and dysraphism [9]. The Ct scan allowed to plan the surgical treatment satisfactorily. Advances in imaging have made it possible to visualize the contents of its appendages in search of an extension of the central nervous system [13,14].

The surgery consisted of the treatment of the meningocele followed by the excision of the appendices at the same time. Intraoperatively, the macroscopic appearance was fibro -adipose. Indeed, on an anatomopathological level, it is made up of adipose, connective, muscular and nervous tissue in variable quantities [15]. As in our cases, the surgery can be simple, the caudal appendage did not contain nerve elements [9]. The postoperative evolution was satisfactory for our patients.

Conclusion

Caudal appendages are markers of dysraphism which can be cranial and caudal. They require a more global assessment using

imaging of the skull and spine associated with an adequate clinical examination. These are rare lesions whose prognosis depends on the presence or absence of associated abnormalities.

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