CASE REPORT

Two cases of polydactyly in wild brown howler monkeys (*Alouatta guariba clamitans*)

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Abstract

We report the first two cases of polydactyly in an atelid species: (i) a wild *ca*. 16-week-old infant female presenting seven digits in both feet and other bone malformations and (ii) a wild newborn male presenting six digits in both feet with the extra digit fused to the hallux.

Introduction

Polydactyly (extra digits on hands and/or feet) is a congenital limb malformation found in primates [2–5, 10, 13, 18, 21, 22, 27], and domestic animals [9]. Whereas polydactylous individuals of some species do not show other external congenital malformations [3, 22], this phenotype may be associated to a syndrome of defects in others [2, 18, 28].

Environmental, genetic, and hereditary factors may cause this malformation [2, 4, 10, 15, 16, 22, 28]. In humans, polydactyly can have genic or chromosomal causes [2, 19, 28], often involving the trisomy of chromosome 13 (HSA13) and, rarely, trisomy 18 [12]. In Old World monkeys, the trisomy for the chromosomal pair homeolog to HSA13 causes clinical features similar to those observed in humans [1, 13, 20, 26].

We report the first two cases of polydactyly in an atelid, the brown howler monkey (*Alouatta guariba clamitans*), a taxon occurring in the southern part of the Atlantic forest in Brazil and northeastern Argentina [7]. We also test whether one of the cases was associated with a trisomy of the chromosomal pair (chromosome 13 or AGU13) with homeology to HSA13 [17].

Humane care guidelines

All research reported in this manuscript met the appropriate national and institutional guidelines for authorized study of wild animals. JCBM held a license (SISBIO n. 23748-1) issued by the Chico Mendes Institute for Biodiversity Conservation/Brazilian Ministry of Environment. The authors adhered to the guide for care and use of experimental animals as promulgated by the American Society of Primatologists Principles for the Ethical Treatment of Nonhuman Primates.

Case study 1

A wild dependent infant female (estimated to be *ca*. 16 weeks old based on body size, weight, and dentition) was rescued from a domestic dog after falling to the ground in a *ca*. 15-ha forest fragment (30°12′03.61′S, 51°00′45.00′W) in Porto Alegre, state of Rio Grande do

Sul, Brazil, on 15 April 2012. She was anesthetized with dextrocetamina hydrochloride (10 mg/kg) for radiographic and myelographic examination and the collection of a 2-ml blood sample from the axillary vein with a heparinized syringe for metaphase obtainment following standard procedures [14]. The chromosome number was

Fig. 1 **(A)** Radiographic image of the lower limbs of the infant female showing the anatomical anomalies: (a) extra digits of the right foot, (b) extra digits of the left foot, (c) phalanges of the left hallux, (d) right tibial diaphysis, and (e) left tibial diaphysis (inserts show pictures of the right and left feet). **(B)** G-banded metaphase of the infant female with the normal karyotype with the chromosomes assembled by morphology, size, and chromosomal G-banding pattern ($2n = 46$, XX) (inset: original metaphase pre-assembly), and **(C)** with numerical chromosomal rearrangements with the chromosomes assembled by morphology, size, and chromosomal G-banding pattern. This metaphase is $2n = 46$, XX, monosomic for AGU11 and trisomic for AGU13 (boxes) (inset: original metaphase pre-assembly). **(D)** Photo of the dead newborn male showing his normal hands and polydactylous feet (insert shows detail of left feet).

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determined from 56 metaphases. The G-banding technique [23] was applied to 40 metaphases for comparison with the taxon's karyotype [17]. The female was raised in captivity until dying on 2 November 2012. Histopathology of lungs and kidneys showed acute interstitial pneumonia, interstitial suppurative nephritis with interstitial necrosis, and pyelonephritis as the possible causes of death.

The female presented normal hands and bilateral preaxial polydactyly (seven digits) in both feet (Fig. 1A). Feet contained an apparently mirror image duplication of fully formed second and third digits. The proximal, middle, and distal phalanges of the extra digits of both feet were similar in shape and size to those of the normal second and third digits. However, their metatarsals were a little shorter (2.0–2.2 vs. 2.4–2.5 cm). The proximal and distal phalanges of the left hallux were fused and shorter (total = 0.8 cm) than those of the right hallux (proximal = 0.9 cm, distal = 0.5 cm). Duplicated cuneiform tarsals were also associated with the extra digits. Other anatomical anomalies included a slightly deformed left tibial diaphysis (length = 4.9 cm, maximum proximal epiphyseal breadth = 1.6 cm, maximum distal epiphyseal breadth = 0.9 cm), a quite deformed right tibial diaphysis shorter than normal (length = 3.5 cm, maximum proximal epiphyseal breadth = 1.4 cm, maximum distal epiphyseal breadth = 1.0 cm) and with a marked inward curvature, a right tibial distal epiphysis with a sequel suggestive of a Salter-Harris fracture, a shorter and deformed right fibula (maximum length = 3.9 cm) compared to the left fibula (maximum length = 5.0 cm; maximum diameter at midshaft was 0.3 cm for both fibulas), and exacerbated bone reaction of the right and left distal femoral diaphysis and metaphysis. All measurements shown above were taken from the radiograph and must be seen as approximations. No trauma was found, and the overall bone density was low. She could move her lower limbs but her feet showed markedly reduced tone. Her prehensile tail also showed low tone and was unable to tightly hold any object or support or to sustain the body's weight.

A modal number of $2n = 46$, XX with 16.1% hypoploid and 3.6% hyperploid cells, and an MI = 4.1% were found. Thirty of the 40 G-banded metaphases (75%) presented a normal karyotype ($2n = 46$, XX, Fig. 1B), whereas the remaining showed random numerical chromosomal rearrangements (Table 1). None $2n = 47$, XX cell exhibited a trisomy for AGU13. Two $2n = 46$, XX cells exhibited a trisomy for AGU13 together with a monosomy for another chromosome pair (one for AGU11 and the other for AGU21; Fig. 1C).

Table 1 Metaphases of the infant female presenting numerical chromosomal rearrangements ($N = 10$).

2*n*	45	46	47
N metaphases	1	6	3
Trisomy	AGU20, AGU22	AGU8, AGU10, AGU11,AGU13, AGU14, AGU22	AGU17, AGU21, AGU22
Monosomy	AGU5, AGU12, AGU17	AGU7, AGU9, AGU19	–
Nullisomy	–	AGU21	–

Case study 2

The corpse of a newborn male with attached umbilical cord was found in the biological collection of the Feliciano Miguel Abdala Private Natural Heritage Reserve (formerly Biological Station of Caratinga, 19°44′S, 41°49′W) in Caratinga, state of Minas Gerais, Brazil. He was found dead in the forest floor by biologist André Hirsch in 1992.

The newborn presented normal hands and bilateral preaxial polydactyly (six digits) in both feet. Externally, the extra digit of each foot appeared to be another hallux fused by the skin to the normal hallux up to the distal portion of their proximal phalanges (Fig. 1D). He was only examined externally.

Discussion

We described the first two cases of polydactyly in the ateline radiation of prehensile-tailed New World monkeys. Previously described congenital anomalies in atelines include spider monkey microdactyly [cited in 3] and male howler monkey polythelia (supernumerary nipples) [24].

The lack of constancy in the chromosome involved in the numerical rearrangements does not support the presence of a chromosomal trisomy as the cause of the female's polydactyly. However, it is not possible to rule out a genic pathology because the resolution level of the employed cytogenetic technique was unable to detect such abnormalities. Additionally, the landscapes where the study sites are immersed make it quite improbable that the cause of either case was maternal exposure to pesticides or other toxic compounds.

Unlike rare exceptions in which polydactylous nonhuman primates were described as healthy [3], the female showed additional anomalies and was unable to tightly hold any object or support with her feet and tail. Therefore, she was unable to sustain her body's weight with them. Difficulty to cling also was reported for a syndactylous (fused digits) *Saguinus nigricollis* [8].

Infants of the semiterrestrial *Macaca fuscata* with malformed limbs and unable to cling survive the first months because of maternal care [16, 25]. The arboreal lifestyle certainly imposed much higher costs to the howler mother than for macaques [16, 25]. Because normal howler infants are carried on the mother's belly during the first month of life [11], we hypothesize that the infant had been capable of clinging to her mother's venter with her hands in early life, especially if the mother helped by holding her against the venter while moving on three limbs as seen in macaques [25]. The task might have become more challenging for both mother and daughter as the latter grew up. However, the infant might have moved to the mother's dorsum as she aged, where she could ride more easily and safely by holding with her hands only. Field data indicate that two- to four-month-old infants spend about half the time in the mother's belly and the other half on her back [11]. Finally, the estimated age at fall coincides with an increase in independence, as time spent independent from the mother reaches about 90% by the fifth month of life [11].

It seems quite reasonable to hypothesize that the newborn male found dead in the forest floor was similarly handicapped. It is possible that he fell from the trees by not being able to firmly cling to his mother's body. Therefore, these two cases of polydactyly concur with the hypothesis that impairment of individual viability partially explains the rarity of congenital malformations in wild populations [6].

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