Received: 29 January 2022 | Revised: 8 May 2022 | Accepted: 8 May 2022

DOI: 10.1111/joa.13693

Journal of Anatomy ANATOMICAL SOCIETY WILEY

ORIGINAL PAPER

Modifications of the locomotor system in habitually quadrupedal humans

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Funding information
National Museum of Natural History

Abstract

The acquisition of habitual bipedal locomotion, which resulted in numerous modifications of the skeleton was a crucial step in hominid evolution. However, our understanding of the inherited skeletal modifications versus those acquired while learning to walk remains limited. We here present data derived from X-rays and CT scans of quadrupedal adult humans and compare the morphology of the vertebral column, pelvis and femur to that of a bipedal brother. We show how a skeleton forged by natural selection for bipedal locomotion is modified when used to walk quadrupedally. The quadrupedal brother is characterised by the absence of femoral obliquity, a very high anteversion angle of the femoral neck, a very high collo-diaphyseal angle and a very reduced lordosis. The differences in the pelvis are more subtle and complex, yet of functional importance. The modification of the ischial spines to an ischial ridge and the perfectly rounded shape of the sacral curvature are two unique features that can be directly attributed to a quadrupedal posture and locomotion. We propose a functional interpretation of these two exceptional modifications. Unexpectedly, the quadrupedal brother and sister show a greater angle of pelvic incidence compared to their bipedal brother, a trait previously shown to increase with learning to walk in bipedal subjects. Moreover, the evolution from an occasional towards a permanent bipedality has given rise to a functional association between the angle of pelvic incidence and the lumbar curvature, with high angles of incidence and greater lumbar curvature promoting stability during bipedal locomotion. The quadrupedal brother and sister with a high angle of incidence and a very reduced lordosis thus show a complete decoupling of this complex functional integration.

KEYWORDS
femur, ischial spine, lumbar curvature, pelvis, quadrupedal humans, sacro-iliac joint, skeletal plasticity, spino-pelvic complex

1 | INTRODUCTION

As establishing bipedal balance is very demanding in the face of gravity, the human skeleton shows different types of modifications associated with bipedal locomotion. Numerous studies in comparative anatomy and human palaeontology have shown that our postcranial skeleton became adapted to habitual bipedalism over the course of millions of years. This has resulted in modifications of heritable traits through natural selection (Lovejoy et al., 1973; Morton, 1952; Schultz, 1930; Waterman, 1929; Wood, 1978). On the other hand, more recent studies have demonstrated that the human postcranial skeleton is also modified during growth in tight association with the acquisition of a bipedal gait through the conjugate action of gravity and muscular forces (Amtmann, 1979; Lovejoy et al., 2000; Tardieu, 1999, 2010; Tardieu et al., 2013).

To assess the nature of the features that develop during growth due to a bipedal stance and the loading of the skeleton during bipedal locomotion, models have been proposed including goats that were deprived of the use of their upper limbs at birth (Slijper, 1942) and young Japanese macaques trained to walk bipedally (Preuschoft et al., 1988). Alternatively, non-walking children can provide a wealth of observations on the development of the skeleton in the absence of the loading induced by a bipedal stance and locomotion. These studies have demonstrated that different postcranial characters, absent in non-walking children, are tightly associated with active bipedal behaviour (Tardieu & Damsin, 1997; Tardieu & Trinkaus, 1994). The study of the same non-walking children after they learned to walk with a walking frame was also instructive (Tardieu & Damsin, 1997; Tardieu & Trinkaus, 1994). However, an exceptional model would be that of an adult human that has never walked bipedally.

In 2005, adult humans who practice habitual quadrupedal locomotion were identified in a family in Turkey (Humphrey et al., 2005). In a village near Iskenderun in southern Turkey, five adults of a family of 19 children suffer from a mutation at a gene of chromosome 17 inducing a loss of a fundamental protein implied in the development of the cerebellum (Ozcelik et al., 2008; Türkmen et al., 2006). The cerebellar hypoplasia results in a drastic lack of balance which renders the acquisition of a bipedal stance and locomotion difficult. Their quadrupedal gait is, however, easy and balanced: it includes the full extension of the knee, the hands are set palm-down, which results in a very inclined vertebral column (Figure 1). Quadrupedal subjects also use a normal sitting position and when they were young attempted to walk bipedally without success, however. The musculoskeletal system of these subjects was, however, never investigated, despite constituting a unique source of information on how a skeleton forged by natural selection for bipedal locomotion is modified when used to walk quadrupedally.

Here, we provide the first data on the postcranial morphology of these quadrupedal walkers to understand the impact of growth on a skeleton which presents the inherited characters of bipedal parents but which has been remodelled by habitual quadrupedal walking. We hypothesise that the modifications associated with bipedal gait acquisition will be absent in the skeleton of these quadrupedal subjects since gravity has not imprinted its mark during the long process of reaching a bipedal balance (Tardieu et al., 2013). Additionally, we asked ourselves how the skeleton became adapted to this unexpected quadrupedal use during growth. Do some features considered as exclusively linked with bipedal locomotion also develop during quadrupedal locomotion?

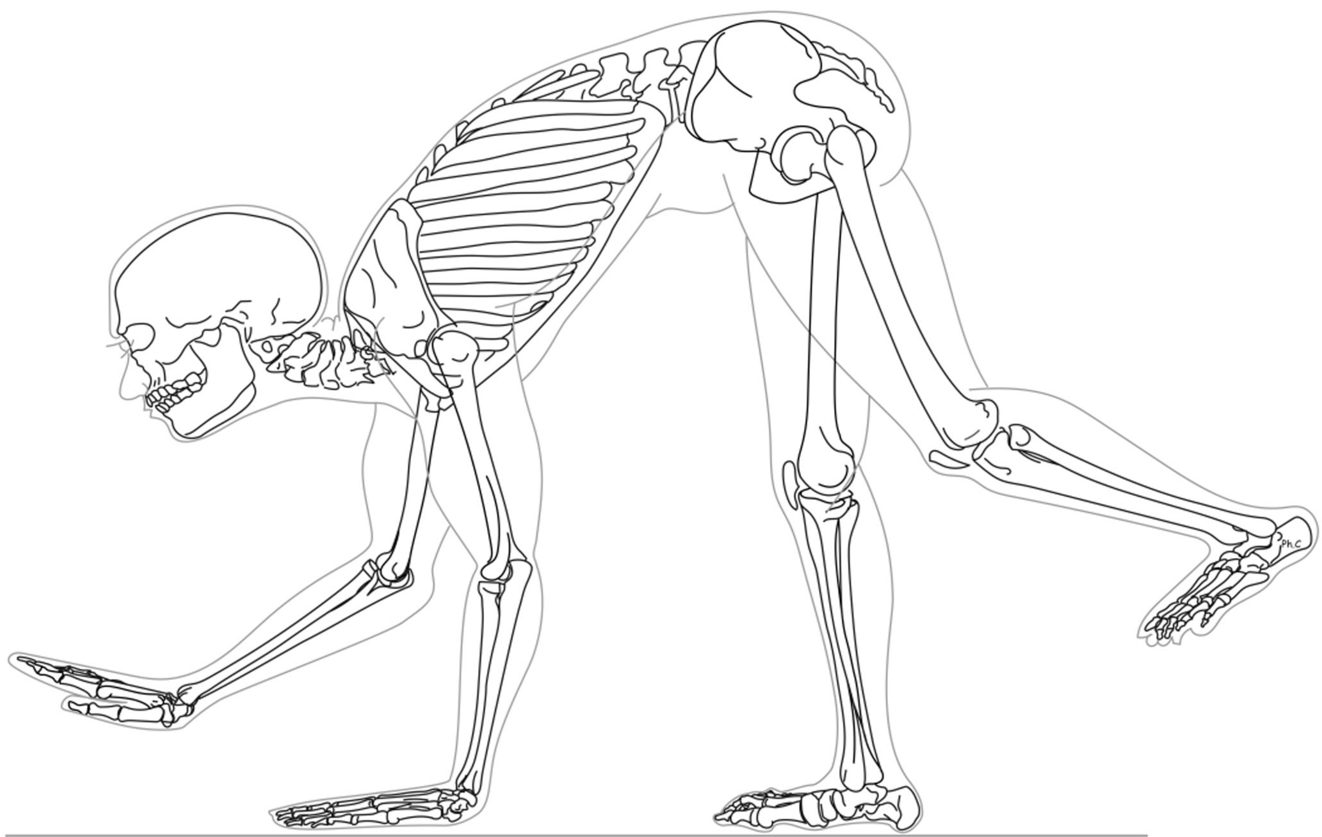

FIGURE 1 Schematic representation of the sister in quadrupedal stance based on the photographs and films taken by Humphrey and collaborators and measurements taken at the hospital in Adana (drawing by P. Campignion)

2 | MATERIALS AND METHODS

2.1 | CT scanning and measurements

Two patients, one 36-year-old man, one 43-year-old woman and a 29-year-old bipedal brothers were examined at the Cukurova University Balcali hospital in Adana, Turkey, where physical examinations, radiographies and CT scanning were performed. CT scans and radiographs were obtained with a medical CT scanner (Toshiba Asteion) at a resolution of 0.708 mm and a slice thickness of 2 mm. CT scans were obtained for the two brothers only and were segmented manually in AVIZO (V. 8, FEI, Hillsboro, Oregon, USA). Measurements (angles, distances) were taken on the CT data in AVIZO or directly on radiographs from the two brothers and one of their quadrupedal sisters following previously published protocols (Tardieu, 2010; Tardieu et al., 2017). The ethics committee at Çukurova University waived the need for ethics approval pertaining to the analysis of these data (request SAYI: 50243401/2019–03) and written consent was obtained allowing the publication of these data. The raw CT data are available from the authors upon reasonable request.

2.2 | Definition of the measurements

Three measurements concern the femur, one measurement concerns the lumbar lordosis of the vertebral column, and two measurements concern the pelvis (Figure 2a,b):

- The angle of femoral obliquity (or bicondylar angle) in adults, measured in anterior view, is the angle between the axis of the femoral diaphysis and the perpendicular to the infracondylar plane (Tardieu, 2010; Tardieu & Trinkaus, 1994).
- The angle of anteversion of the femoral neck, measured in superior view, is the angle between the axis of the femoral neck and head and the infracondylar plane (Martin & Saller, 1957). It measures the anterior torsion of the head and neck in relation to the infracondylar plane. On axial slices of C.T. scans, it is often referred to as the angle between the axis of the femoral neck and the posterior condylar axis.
- The collo-diaphyseal angle (or neck-shaft angle), measured in anterior view, is the angle between the axis of the head and neck and the axis of the diaphysis (Martin & Saller, 1957; Tardieu, 2010).
- The lumbar lordosis is measured on x-rays in lateral view between the axis of the superior endplate of the first lumbar vertebra and of the first sacral vertebra (Boulay et al., 2006; Legaye et al., 1993). For the two quadrupedal subjects, it was measured while in bipedal stance with the subjects being assisted by the operator.
- The angle of pelvic incidence, measured in lateral view, is the angle between the sacro-acetabular distance and the line perpendicular to the superior surface of the first sacral vertebra at its centre. The sacro-acebular distance is the line connecting this centre to the middle of the interacetabular distance (Legaye et al., 1993, 1998). This angle was measured on the CT scans except for the quadrupedal sister for which it was measured on the x-rays.
- The sacral slope, measured in lateral view, is the angle of inclination of the superior endplate of the first lumbar vertebra in relation to the horizontal. As it is a positional parameter depending upon the position of the subject, it is measured in relation to a standard reference plane of erect position, called the Lewinneck plane, determined by the two superior iliac spines and the superior extremity of the pubic symphysis (Lewinneck et al., 1978). The sacral slope of the pelvis quadrupedal sister was measured on x-rays with verification of the vertical position of her pelvis.

2.3 | Comparison of the two pelves

To compare the pelves of the two brothers, we scaled the structures based on their centroid size. Centroid size was calculated by placing 18 landmarks distributed across the entire object. Objects were scaled in AVIZO (V.8). The scaled pelves were then superimposed using a least squares best fit algorithm in Geomagic Studio (V.12). Videos showing the skeletal elements of each of the brothers (Supplementary videos) were made using Blender (V2.78c, Blender Foundation).

3 | RESULTS AND DISCUSSION

3.1 | Morphology of the femur

We measured the angle of femoral obliquity and the two angles that quantify the orientation of the femoral neck: the anteversion angle of the neck and the collo-diaphyseal angle (Table 1 and Figures 2 and 3). The obliquity of the femoral shaft equals 7° on both the right and left sides in the bipedal brother. This angle is zero on both sides in the quadrupedal brother (see also Videos S1 and S2). The mean angle of femoral obliquity in normal adults ranges between 8° and 11° (Lovejoy, 1978; Tardieu, 1983). The obliquity of the femur positions the knee close to the line of gravity of the body during the single stance phase of walking, thus contributing to the efficiency of the human bipedal striding gait. This angle is 0° at birth and its increase closely parallels the acquisition of walking in young children and reaches its final values around the age of 8 years (Tardieu & Trinkaus, 1994). We previously showed that femoral obliquity does not develop in non-walking children (Tardieu, 2010; Tardieu & Damsin, 1997). This angle develops through a medial metaphyseal apposition at the distal end of the femoral physis during longitudinal diaphyseal growth (Pauwels, 1979; Shefelbine et al., 2002; Tardieu, 2010). This differential mediolateral metaphyseal apposition is possible only because the human distal physis is flat at birth and remains flat during growth (Tardieu & Preuschoft, 1996). At the opposite, very early in the development of non-human primates, the

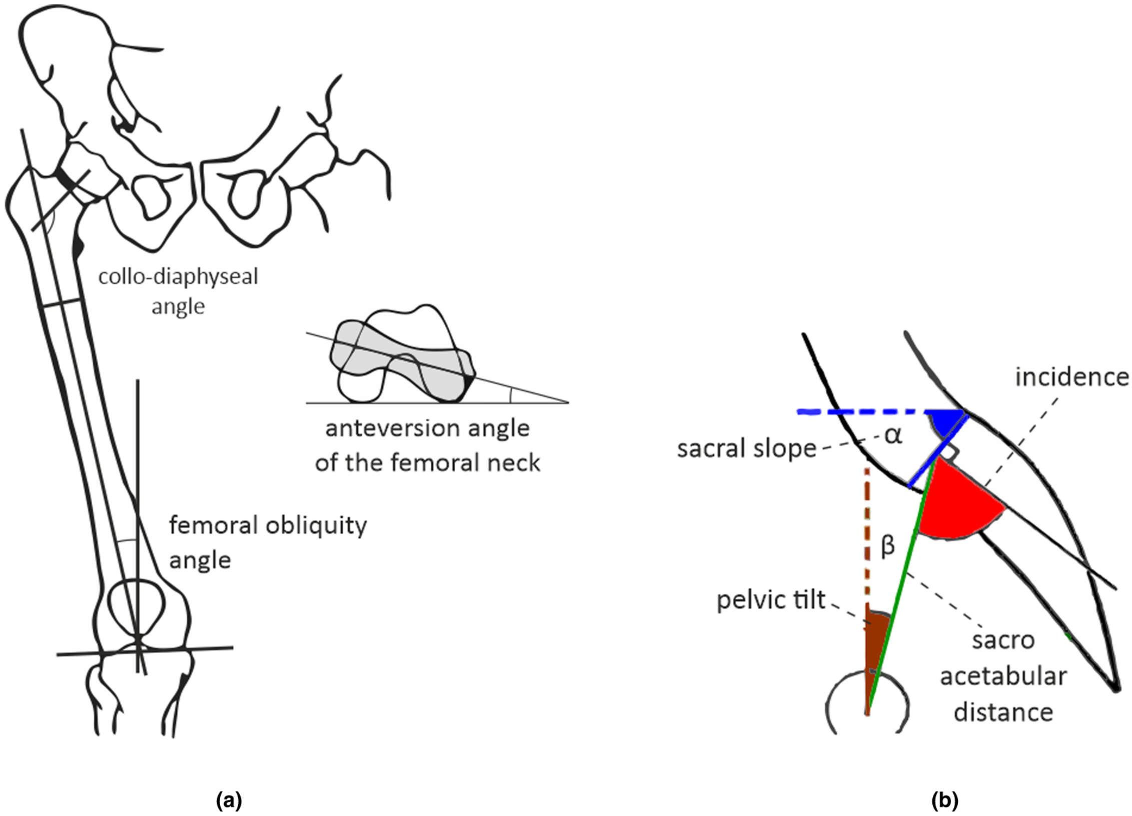

FIGURE 2 (a) Femoral measurements. Bicondylar angle, neck-shaft angle and angle of anteversion of the neck. (b) Lumbar and pelvic measurements in lateral view. The two acetabula are superimposed. The angle of pelvic incidence is defined by the sacro-acetabular distance -linking the centre of the superior surface of the first sacral vertebra and the middle of the biacetabular distance- and the perpendicular to the centre of the superior surface of the first vertebra. This angle is the geometric sum of the two positional variables, sacral slope and pelvic tilt

TABLE 1 Values of the six studied parameters with the mean values and standard deviations for representative human samples. A comparison with a sample of Turkish subjects is also provided for the anteversion angle of the neck and the cervico-diaphyseal angle (second value for the parameter). L, left; R, right

	Normal brother		Quadrupedal brother				
	R	L	R	L	Quadrupedal sister	Means ± S.D.	*N*
Angle of femoral obliquity	7°	7°	0°	0°		8° ± 1.6° 11° ± 1.3°	73 50
Anteversion angle of the neck	14°	12°	28°	24°		11.5° ± 9° 13.1° ± 8° 12.5° ± 8°	15 85 100
Cervico-diaphyseal angle	127°	129°	140°	136°		125° ± 4° 130.6° ± 7° 130° ± 6°	51 85 66
Lumbar lordosis	55°		32°		22°	55° ± 11.2° 66° ± 9.4°	126 149
Pelvic incidence	41°		57°		67°	51° ± 10.9° 55° ± 10.6°	160 300
Sacral slope	34°		40°		25°	41° ± 14°	51

distal physis is divided by two grooves, each corresponding to a crest on the distal epiphysis. This complex fit of the epiphysis into the diaphysis is required to prevent epiphyseal separation in the context of an arboreal mode of life (Tardieu, 1993, 2010). Two unfused femoral diaphyses of australopithecines with a high angle of obliquity show that 3 million years ago this complex fit had already changed and evolved into the simplified form typical of humans (Tardieu, 2010). This modification was selected in the context of human bipedal

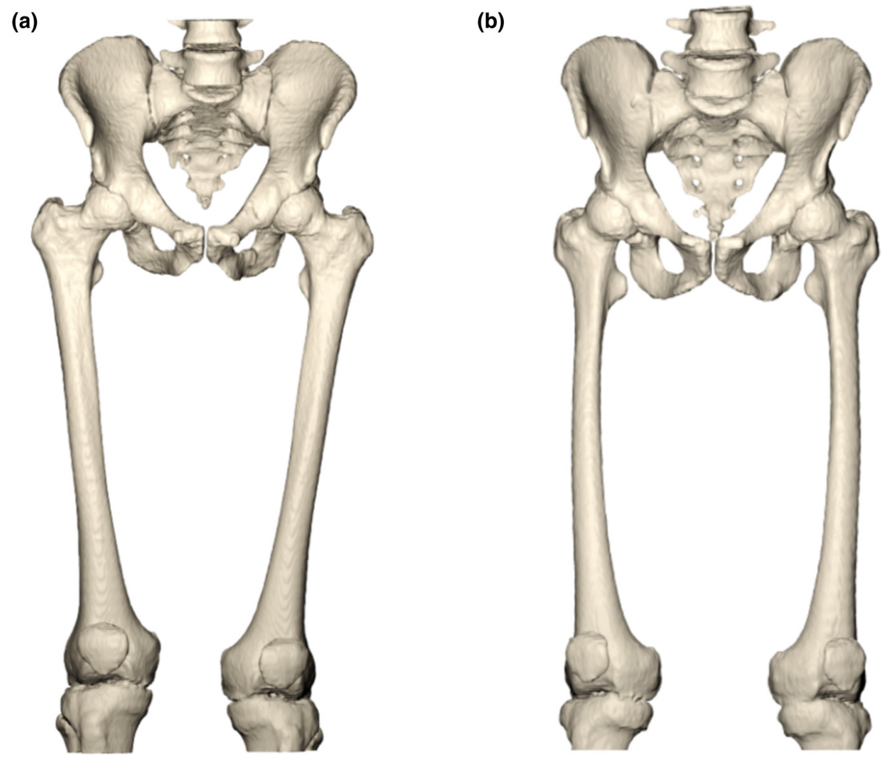

FIGURE 3 Three-dimensional reconstruction of the pelvis and femur of the two brothers. (a) Frontal view of the bipedal brother. (b) Frontal view of the quadrupedal brother. The absence of femoral obliquity in the quadrupedal brother is clear. The high anteversion of the femoral neck is manifest through the medial visibility of the lesser trochanter, the lateral position of the patella and the weak coverage of the femoral head. In this supine position from CT-scan the femur is placed in external rotation relative to the acetabulum

locomotion and is inherited. The formation of the bicondylar angle was necessarily preceded by this modification, which suggests a link between the two processes, natural selection and loading of the skeleton during bipedal locomotion. We observed that the femur of the quadrupedal subject did not develop an obliquity, in spite of the presence of a flat distal physis on his femur at birth. We can thus conclude that the formation of the bicondylar angle is exclusively linked with the practice of bipedal locomotion.

The mean anteversion angle of the femoral neck is between 11.5° and 13° in the adult human population (Biçer et al., 2016; Reikeras, 1991; Von Lantz, 1953). A measurement of 14° and 12° for the right and left femoral neck anteversion was obtained for the bipedal brother. The values obtained for the quadrupedal subject were greater, as 28° and 24° were measured on the right and left femora respectively. During prenatal growth the torsion of the femoral shaft increases which results in an increase in the angle of anteversion of the femoral neck (Bonneau et al., 2011; Fabry et al., 1973). The progressive torsion in utero is produced by mechanical constraints induced by the contention of the foetus in the uterine cavity (Bonneau et al., 2011). The mean values of the anteversion angle at birth is around 35° and this angle decreases during postnatal growth (Svennigsen et al., 1989). This decrease is associated with the progressive acquisition of the full extension of the hip joint by the young child, which occurs quite late in development with the acquisition of a bipedal gait. The resistance of the pretrochanteric bundle of the ilio-femoral ligament at the anterior part of the femoral neck, counteracting the force of the hip extensors towards the full extension of the hip, results in the progressive rotation of the superior epiphyseal block in relation to the diaphysis (Le Damany, 1905). We observed that the decrease in the angle of anteversion occurred normally in the bipedal brother. At the opposite, we suggest that the decrease did not occur during the growth of the quadrupedal brother because he never used full hip extension. In bipedal humans, to compensate a high neck anteversion the femur must be placed in internal rotation during bipedal walking so as to increase the articular congruency between femoral head and acetabulum (Tardieu, 2010). In the quadrupedal brother, the flexion of the trunk makes the amplitude of this internal rotation even greater, which likely involves the strong solicitation of the external rotator muscles. This situation has important consequences on the morphology of the ischial region of the pelvis.

The mean femoral collo-diaphyseal angle is on average 125°–130° in humans (Biçer et al., 2016; Parsons, 1914; Reikeras et al., 1982). Angles of 127° and 129° were measured on the right and left femora of the bipedal brother while angles of 140° and 136° were measured for the femurs of the quadrupedal brother. In foetuses and new-borns, a defined neck is absent. During growth, the neck-shaft angle decreases from 145° to 125° (Heimkes et al., 1993; Reikeras et al., 1982). We previously showed that in non-walking children, this angle does not decrease (Tardieu, 2010). Moreover, a progressive increase in this angle is observed after excision of the hip abductor muscles (Brien et al., 1995). The decrease and increase in this angle are related to the same mechanisms of differential metaphyseal apposition described for the femoral bicondylar angle. We observe that in the bipedal subject this angle decreased during growth to reach a normal adult value. In contrast, in the quadrupedal subject, this angle did not decrease or only weakly because of the absence of a bipedal loading of his hip joints.

3.2 | Morphology of the vertebral column

We measured the degree of lumbar lordosis of the vertebral column (Table 1). The mean angles of lordosis in human populations range from 55° (Tardieu et al., 2017) to 66° (Boulay et al., 2006). The normal brother had a lordosis of 55°, while the quadrupedal brother only reached 32° and the quadrupedal sister only 22°. We measured the sacral slope of the two brothers in reference to the plane of Lewinneck. The sacral slope of the quadrupedal brother is 40°, the slope of the bipedal brother is 34° and the one of the quadrupedal sister as measured on x-rays, is 25°. The mean sacral slope in newborns is 24° (±16.6°) and reaches 40.9° (±8.4°) in adults (Tardieu et al., 2013, 2017). The graph which illustrates the relationships between lordosis and sacral slope (Figure 4a) shows that the two quadrupedal subjects deviate from the normal values due to their very low lordosis while the bipedal brother falls within the normal range. In bipedal humans, the angle of lordosis is positively correlated with an important pelvic sagittal parameter, the angle of pelvic incidence (Figure 4b). These two parameters are crucial in the acquisition of the sagittal balance of the trunk on the lower limbs and consequently in the comparison between bipedal and quadrupedal subjects.

3.3 | Morphology of the pelvis

We compared the pelvis morphology of the two brothers by superimposition of the two pelves (Figure 5a). Noticeable differences are the slightly stronger curvature of the iliac blades and the stronger curvature of the distal portion of the sacrum of the quadrupedal brother (see Videos S3 and S4). At birth, there is no sacral curvature on the anterior surface of the sacrum. The sacral curvature is a plastic character since a supine sleeping posture causes an increase in the sacral curvature while a prone posture results in its decreases in young children (Abitbol, 1989). During the process of trunk straightening, the superior part of the sacrum becomes more horizontal: the sacro-lumbar muscles (*multifidus* and *iliocostalis lumborum*) pull the dorsal part of the sacrum backwards at the level of the first and second sacral vertebrae. The sacrum curves under the distal tension of the powerful sacrospinous ligaments and of the ischio-coccygeal muscles (Tardieu et al., 2013). The lateral view of the pelvis of the bipedal brother illustrates this process (Figure 5b): the two first sacral vertebrae were pulled more horizontally as demonstrated by their well-developed dorsal spines. Then the distal curve becomes more

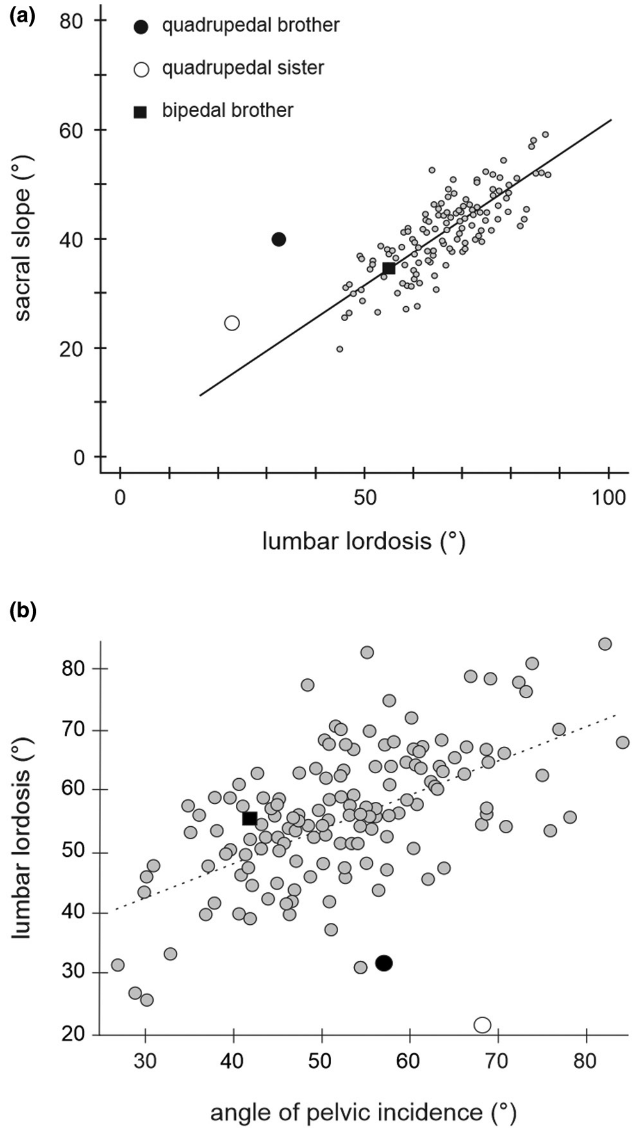

FIGURE 4 (a) Relationship between sacral slope and lumbar lordosis in a sample of 147 normal healthy subjects ($r = 0.80$; $p < 0.001$) (Boulay et al., 2006). (b) Relationship between the angle of pelvic incidence and lumbar lordosis in 131 normal subjects ($r = 0.55$; $p < 0.001$) (Tardieu et al., 2017). Data were obtained from x-rays. The black square represents the bipedal brother, the black circle the quadrupedal brother and the white circle the quadrupedal sister

pronounced at the level of the last sacral vertebrae. In contrast to this sagittal sacral shape including two distinct parts, the sacrum of the quadrupedal brother (Figure 5c) presents a perfectly regular and continuous circular shape; the dorsal spines of the two first sacral vertebrae are only weakly developed, which is in accordance with the weak lordosis. This circular shape of the sacrum in lateral view, which signals the absence of trunk erection, could be considered as a signature of quadrupedal locomotion in a human subject.

The comparison also shows that the ischial spines of the quadrupedal brother are located at a lower level on the ischium and present a distally extended area of insertion (Figure 5c). The ischial spines appear in early adolescence (Abitbol, 1988). The superior gemelli

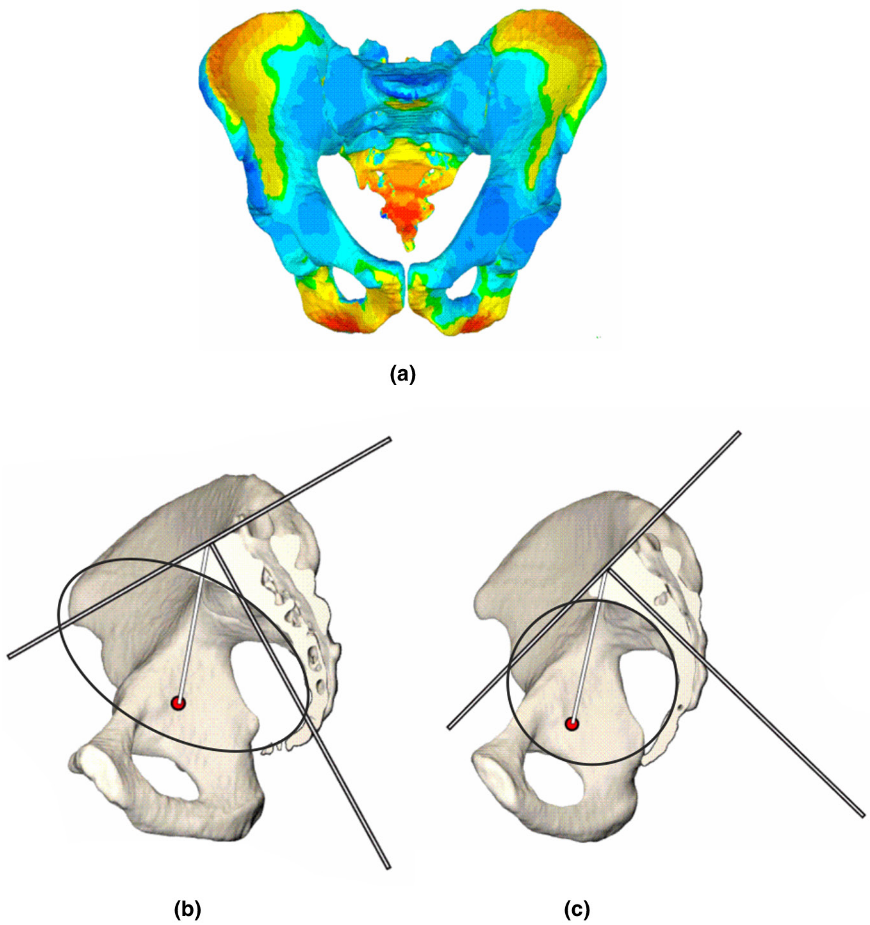

FIGURE 5 Comparison of the pelves of the two brothers. (a) Frontal view: Pelves of the two brothers are scaled and superimposed. Differences are illustrated using a colour scale. Hotter colours illustrate greater differences between the two pelves. Note the difference in the position of the ilia which are curved inward in the quadrupedal brother. (b, c) lateral view: Pelves not scaled. (b) Virtual hemi-section of the pelvis of the bipedal brother. (c) Virtual hemi-section of the pelvis of the quadrupedal brother. Note the angle of pelvic incidence which is larger in the quadrupedal brother, the shape of the sacral curvature which is almost circular in the quadrupedal brother but elliptic in the bipedal brother, the weak dorsal spines in the quadrupedal brother but the robust ones in the bipedal brother and the shape of the ischial spine which is extended distally in the quadrupedal brother

which insert in the trochanteric fossa originate from the ischial spines. Arising from the same femoral insertion, the inferior gemelli originate close to them, distally from the ischium. These two muscles insert onto the tendon of the obturator externus, which has the same femoral insertion and is a more powerful muscle than the two gemelli (Figure 6a,b). These three muscles are external rotators of the femur. Our hypothesis is that because of the high anteversion of the femoral neck of the quadrupedal subject these muscles are strongly solicited during quadrupedal stance (Figure 6b). The continuous conjoint contraction of these muscles would have resulted in the formation of bone at the level of the ischial origin of the superior gemelli and finally in the formation of an enlarged ischial ridge of origin instead of the unique and well-differentiated ischial spine. The sacro-spinous ligament, which inserts on the ischial spine and on the lateral upper distal half of the sacrum, is likely less solicited in the quadrupedal stance, as reflected in the absence of a marked protuberance of the spine. This is also reflected by the perfectly round shape of the sacrum revealing the absence of an opposite displacement between the proximal and distal parts of the sacrum during growth. We propose here that the remodelling of the skeleton of the quadrupedal subjects results from the particular way their muscles work and not from the passive action of their ligaments only. Figure five (a, b, c) shows that the ischio-pubic ramus and the ischiatic tuberosity are far less robust in the quadrupedal subject than in his brother. This suggests that the recruitment of the semitendinosus muscle, of the long head of the biceps femoris and of the adductor magnus during locomotion is greater in the bipedal brother than in the quadrupedal one.

3.4 | Sagittal relationships between pelvis and spine

We measured the angle of pelvic incidence (Figures 2 and 7; Table 1), a sagittal parameter which describes the location of the sacrum in relation to the acetabula and which is tightly and positively correlated with the degree of lumbar lordosis in adult

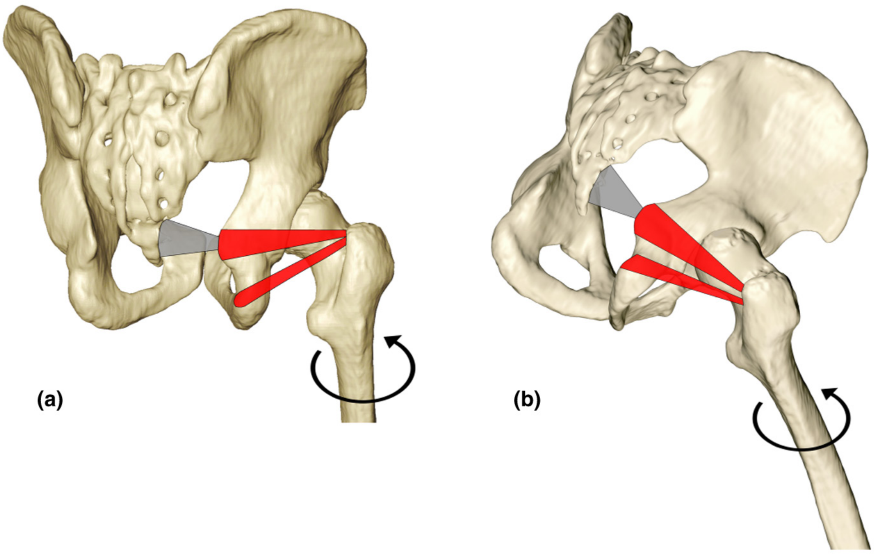

FIGURE 6 Pelvis and upper femur of the quadrupedal brother. (a) Postero-lateral view in erect position. (b) Postero-lateral view in quadrupedal stance. The position of the superior and inferior gemelli muscles and the sacro-spinous ligament are described. The anterior tilt of the pelvis in quadrupedal stance increases the tension of the two muscles which are then stretched and contributes to the enlargement of the ischial spines. The space between the two ischial insertions of the gemelli shows the area where the internal obturator muscle passes across the ischium creating a pulley effect. The arrow indicates the internal rotation of the femur during walking due to the strong anteversion of the femoral neck; this permits to increase the congruency between the femoral head and the acetabulum

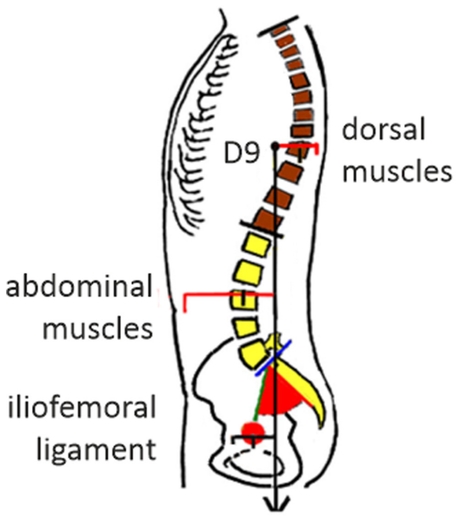

FIGURE 7 Functional link between pelvis and spine in lateral view. Economic sagittal balance of the trunk on the lower limbs, based on the experimental work of Duval-Beaupère and colleagues (1992). The Centre of gravity of the trunk is usually located in front of the 9th dorsal vertebra. The line of gravity of the trunk is anterior to the dorsal curvature, posterior to the lumbar curvature and posterior to the hip joints. On this subject the value of the lordosis is 63°, the value of the pelvic angle, called pelvic incidence, is 64°. These two values are located on the line of regression of the positive correlation between these two parameters presented in Figure 4b

humans (Duval-Beaupere et al., 1992; Legaye et al., 1993; Legaye et al., 1998; Tardieu et al., 2017). Its importance is provided by the fact that this parameter represents the sum of two positional parameters: sacral slope and pelvic tilt (Boulay et al., 2006; Legaye et al., 1993). It is the signature of a subject allowing, in association with the degree of lumbar lordosis, the evaluation of the efficiency of sagittal balance. The mean angle of incidence in newborns is $27° \pm 12°$ (Tardieu et al., 2013). The mean angles of incidence in adults ranges from $51° \pm 10.9°$ to $55° \pm 10.6°$ (Berthonnaud et al., 2005; Vialle et al., 2005). The angle of pelvic incidence of the bipedal brother is 41°, the one of the quadrupedal brother is 57° and the one of the quadrupedal sister is around 67°. The high values of the angle of pelvic incidence in the quadrupedal subjects (57° and 67°) were an unexpected result. We expected low values for the pelvic incidence because these subjects never adopted a bipedal stance or locomotion. Indeed, the increase in the angle of pelvic incidence during human growth has been well documented and is considered as being tightly associated with bipedal gait acquisition (Tardieu et al., 2013; Thiong et al., 2007). The increase in incidence gives stability to the superior surface of the body of the first sacral vertebra for the deployment of the vertebral curvature (Figure 8a). Furthermore, it has been suggested that during the transition from an occasional to more permanent bipedalism in hominid evolution, the pelvic incidence increased at the same time as the sacro-acetabular distance decreased (Figure 8b). This would have then resulted in the creation of the correlation between this angle and the degree of lordosis. From that point onwards, the pelvis and spine became a single functional unit in hominin evolution (Tardieu et al., 2013, 2017; Tardieu & Hauesler, 2019). This complex link, a heritage of natural selection, is established during growth through the loading of the skeleton by gait acquisition and is achieved in adulthood (Tardieu & Hauesler, 2019). However, here we observe a strong contrast between the low lordosis and the high value of pelvic incidence in these quadrupedal subjects.

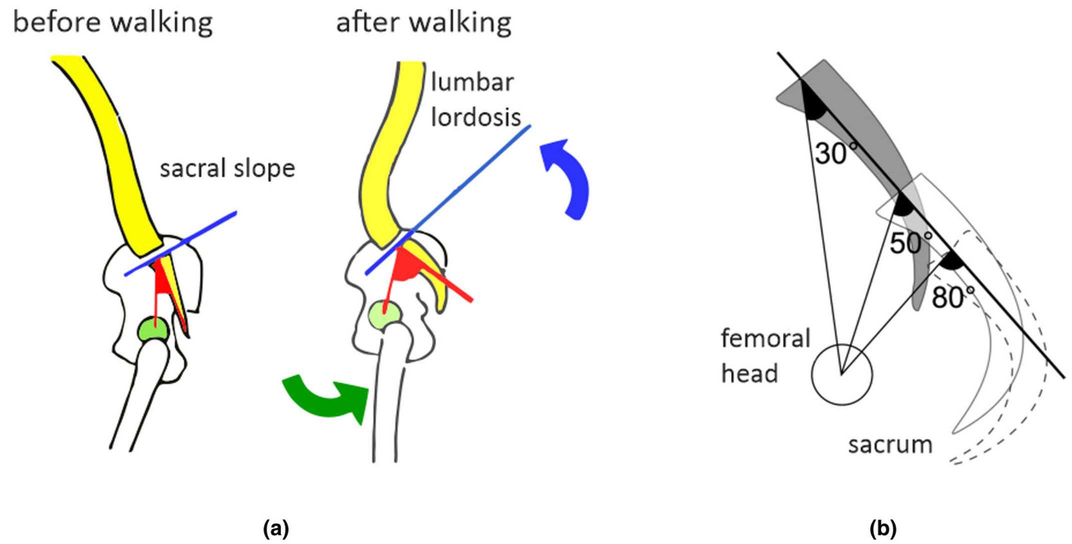

FIGURE 8 Ontogenetic and phylogenetic model of the increase in the angle of pelvic incidence. (a) Pelvic mechanism of trunk strengthening. Skeletal modifications: On the left: Before walking, hip flexion and anterior vertebral flexion induce an anterior location of the trunk centre of gravity. On the right: After walking, femoral extension, lumbar curvature, increase in sacral slope and pelvic incidence entail a backward displacement of the trunk centre of gravity. The backward displacement of the sacrum in relation to the acetabula is visible (for muscular actions in trunk straightening, see Tardieu et al., 2013, Figure 8b). (b) Sagittal model of the opposite relationships between the angle of pelvic incidence and the sacro-acetabular distance. Three schematic steps are depicted: A theorical ancestral stage (30°), an australopithecine stage (50°) not far from the human mean (54°) and and extreme range for humans (80°). This figure is schematic because the sacral slope is kept constant to emphasise the shortening of the sacro-acetabular distance with increasing pelvic incidence. This model illustrates the more and more backward positioning of the sacral plate in relation to the acetabula, a key factor of the sagittal bipedal balance in humans, promoting a backward displacement of the trunk centre of gravity over the lower limbs

The correlation between these two parameters lies beyond the typical variation (Figure 4b). This difference illustrates the total disconnect between pelvic incidence and lordosis in the quadrupedal subjects: during growth, the angle of pelvic incidence increases strongly while lordosis does not develop. The functional association between spine and pelvis is ruptured.

How to explain the high value of the angle of pelvic incidence in the quadrupedal subjects? Two suggestions are possible. The first is based on the radiographic observation of the profile view of the lumbar column of the quadrupedal brother. His weak lumbar curvature is very atypical. The superior and inferior plates of the four first lumbar vertebrae are almost horizontal and their anterior borders are almost vertical. The curvature is restricted to the most distal part of the lumbar column: from the inferior plate of the fourth vertebra, still horizontal, to the very inclined superior plate of the first sacral vertebra. The wedging of the fifth lumbar vertebra initiates the lordosis. It is separated from the first sacral vertebra by a large inter-articular space which is very opened anteriorly. These features draw a clear lordotic curvature but are concentrated on three very distal lumbar vertebrae. We suggest that the frequent sitting posture of the quadrupedal subjects may have played an important role in the modelling of this particular lordotic curvature. It includes a strong inclination of the superior plate of the first sacral vertebra which is an important component of the increase in the angle of pelvic incidence (Figures 2, 7 and 8a). The sitting position includes a clear erection of the trunk without anterior thoracic flexion.

On the other hand, the increase in the angle of incidence can be due to the change in the orientation of the sacro-iliac joints, either during prenatal or postnatal growth: a rotation occurs between the sacrum and the ilia, with the sacral plate rotating anteriorly. This rotation may occur at the end of prenatal growth with intra-uterine constraints (Tardieu et al., 2017) and occurs during postnatal growth with gait acquisition. When the joints are still cartilaginous in infants and young children, the mobilisation of the sacro-iliac joints is facilitated. To increase the angle of incidence through the action of sacro-iliac joints, a couple of opposite forces must apply to the sacrum on the one hand and on the ilia on the other hand (Figure 9). During quadrupedal walking, the viscera are no longer supported by

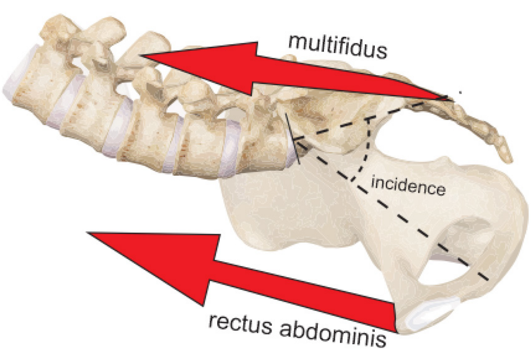

FIGURE 9 A hypothesis for the high incidence in the quadrupedal subjects. Schematic illustration of the pelvis and part of the vertebral column illustrating the couple of muscle forces (*multifidus* and *rectus abdominis*) acting to increase the incidence in the quadrupedal subjects. Notice that the sacral slope in quadrupedal posture is always located beyond the vertical position

the bottom of the pelvis as in bipedal subjects. Consequently, the abdominal muscle (*rectus abdominis*) originating from the pubic symphysis must be active. This could then constitute the force which rotates the pubis anteriorly and the ilia clockwise. Quadrupedal progression likely also mobilises the dorsal musculature inserted on the sacrum to a great degree. For example the spondylolysis on the fifth lumbar vertebra in the quadrupedal subject is indicative, and may even be symptomatic, of this strong mobilisation. The concerned muscles are the distal part of the *multifidus* originating on the five lumbar vertebrae and the most medial bundles of the erector spinae, its *spinalis* part originating on the fifth lumbar vertebra. They would represent the forces that tend to rotate the sacrum anticlockwise around the sacro-iliac joints: this sacral nutation increases the sacral slope. As a hypothesis, we suggest that this couple of forces could be responsible for the increase in the angle of pelvic incidence in the infant or very young quadrupedal subjects. The sacro-iliac joints of humans thus prove to be key-joints whose mobility would be unavoidable irrespective of the mode of locomotion. The high values of this angle would no longer be considered as being exclusively linked with a practice of habitual bipedalism but rather as a response to the specific loading conditions during different locomotor modes. The angle of pelvic incidence thus would acquire the status of a more generalised parameter than previously considered.

Despite the significant insights into the changes in the morphology of the vertebral column, pelvis and femur of adult humans that have never walked upright, our study is clearly limited by the low sample size and the lack of data on the kinematics and dynamics of locomotion in the quadrupedal subjects. Information on the hind limb muscles through MRI would also be important to better understand the observed osteological modifications in the pelvis and hind limb.

4 | CONCLUSION

The morphology of the locomotor skeleton of the quadrupedal brother is different from that of his bipedal brother. Our results demonstrate marked differences in the femur, pelvis and lumbar vertebrae. Differences in the femur are the lack of obliquity of the femur and the absence of closure of the neck-shaft angle and of the anteversion of the femoral neck in the quadrupedal brother. The differences in the pelvis are more subtle and complex, yet of functional importance. The modification of the ischial spine to an ischial ridge and the perfectly round shape of the sacral curvature are two unique features that can be directly attributed to a quadrupedal posture and locomotion. In bipedal humans, as a result of a long evolutionary process, the angles of pelvic incidence and lumbar curvature have become positively correlated, as observed in the bipedal brother. In the quadrupedal brother and sister, we observe a decoupling of this integration between pelvis and lumbar column: the marked increase in the angle of pelvic incidence during growth is associated with a near-absent lumbar curvature. The increase in the angle of pelvic incidence which is tightly associated with learning to walk in bipedal humans was therefore unexpected in the quadrupedal brother and sister. Our data provide novel insights into the plasticity of the human hind limb skeleton and the modifications occurring in a skeleton adapted for bipedal locomotion when adopting a habitual quadrupedal posture and locomotion. This has resulted, most notably, in the rupture of the functional association between the pelvis and vertebral column and the absence of femoral obliquity.

ACKNOWLEDGEMENTS

We thank Sevket Sen, research director at the CNRS and palaeontologist in the National Museum of Natural History of Paris who gave us advice and helped us prepare the project and the two trips to Turkey. We thank the radiologists and orthopaedic assistants who performed the scans and x-rays of the patients in the Hospital of Adana. We thank P. Campignion, physiotherapist and teacher of physiotherapy, who used his anatomical knowledge to reconstruct the skeleton of the sister in a quadrupedal stance.

AUTHORS' CONTRIBUTIONS

C.T. conceived the manuscript; O.D., E.A., L.O., O.S.B., C.T. and AH collected data; C.T., A.D., R.C. and A.H. analysed the data; C.T. and A.H. drafted the manuscript and all authors reviewed the manuscript.

DATA AVAILABILITY STATEMENT

Data are available from the authors upon reasonable request.

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How to cite this article: Tardieu, C., Demirhan, O., Akbal, E., Ozgozen, L., Biçer, Ö.S., Delapré, A. et al. (2022) Modifications of the locomotor system in habitually quadrupedal humans. *Journal of Anatomy*, 00, 1–11. Available from: https://doi.org/10.1111/joa.13693