CASE REPORT

Mary S. Megyesi,¹ M.S.; Ryan M. Tubbs,^{1,2} M.A.; and Norman J. Sauer,¹ Ph.D., D.A.B.F.A.

An Analysis of Human Skeletal Remains with Cerebral Palsy: Associated Skeletal Age Delay and Dental Pathologies*

ABSTRACT: In 2002 the authors were asked to examine the skeletal remains of an individual with a known history of severe cerebral palsy (CP) who was 21–23 years old at death. Skeletal age estimates of 11–15 years and dental age estimates of *c*. 16 years are younger than the known age of the decedent. Skeletal analysis also identified dental pathologies such as chronic tooth grinding and substantial calculus deposits. Scarce literature exists on forensic human remains cases with CP, and this study contrasts the age discrepancy and other features of this case with typical clinical characteristics of CP. A review of the CP literature suggests that delayed skeletal maturation and dental pathologies such as those observed in this case are indicative of complications related to CP. This article may alert future investigators to some of the osteological signs of CP and the probability that age indicators may be misleading.

KEYWORDS: forensic science, forensic anthropology, cerebral palsy, epiphyseal fusion, skeletal development, age estimation

Cerebral palsy (CP) describes a group of disorders of movement and posture causing activity limitation that are nonprogressive and attributed to neurological disorders during fetal or infant development (1). The United Cerebral Palsy (UCP) foundation estimates that nearly 800,000 children and adults in the United States are living with one or more of the symptoms of CP (http://www.upc. org/).

The purpose of this analysis is to add to the scarce literature on forensic human remains cases with CP and to examine and discuss some of the unusual skeletal and dental characteristics of this case. The degree of epiphyseal fusion is delayed with respect to chronological age and dental development. In addition, there are atypical dental pathologies that are consistent with complications of CP. Documenting these characteristics in one individual with CP can reveal how some aspects of CP may be evident in skeletal remains.

Case Background

In October of 2002 Saginaw and Michigan State Police recovered human remains from a residence near Saginaw, MI. The Michigan State University Forensic Anthropology Laboratory was asked to determine the biological profile, assist with evaluating a positive ID, and analyze the remains for evidence of trauma. The remains were completely skeletonized and all remaining soft tissues were in an advanced decompositional state consisting of only liquids and semi-solids. It was immediately apparent upon initial examination that the skeleton was unusual, gracile, and undersized.

¹Department of Anthropology, Michigan State University, 354 Baker Hall, East Lansing, MI 48824.

²College of Human Medicine and Department of Radiology, Michigan State University, A134 Life Sciences, East Lansing, MI 48824.

*A portion of this research was presented at the 57th Annual Meeting of the American Academy of Forensic Sciences in New Orleans, LA, February 21–26, 2005.

Received 23 Jan. 2008; and in revised form 15 April 2008; accepted 20 April 2008.

Strong circumstantial evidence gathered during the police investigation of this case including medical records, evidence gathered from the residence, depositions of family members, and other correlating material from the skeletal remains resulted in an identification of the remains as an African-American male diagnosed with severe CP who was between 21 and 23 years old[†] at death.

Skeletal Analysis

The skeletal remains represented one nearly complete individual, missing only a few small hand and foot bones. Skeletal elements such as the sacrum and tarsals were noticeably light and fragile and the skeleton was extremely gracile. For example, the maximum midshaft diameter of the right femur was 20 mm. Figure 1 shows the right femur of our case compared with that of an average-sized adult middle-aged white male from the MSU Forensic Lab Collection to illustrate the extreme size difference. No perimortem, postmortem, or antemortem trauma was found on the skeleton. African-American ancestral traits were noted in the mid-face such as nasal guttering, wide nasal aperture, and prognathism. Sex was

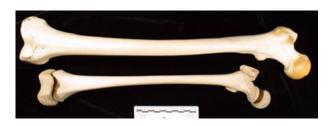


FIG. 1—Comparison of CP case femur (smaller with open epiphyses) with an average adult male femur.

[†]The exact length of time between death and when the individual was found and recovered is unknown but could not exceed 2 years. Therefore, the minimum chronological age for this individual is 21 years and the maximum is 23 years.

not determined skeletally due to the immaturity of the remains and as the individual was known to be male.

Skeletal age determination was an important consideration in this case in order to create a biological profile and verify the identity of the decedent. However, there were a number of inconsistent aging indicators within the skeleton that made determining age difficult. We also had to consider the implications of the extreme gracility and overall immaturity of the skeleton. Were these skeletal characteristics consistent with the symptoms and complications of CP? Delayed skeletal maturation and dental pathologies in this individual are unusual, but are consistent with much of the clinical literature documenting the disabilities, symptoms, and conditions of individuals with CP. The next section describes the skeletal and dental age estimates and the dental pathologies in this case.

Skeletal Analysis: Age Estimation

Skeletal age estimates based on the timing of epiphyseal fusion were determined for males as sex was known for this individual. The remains appeared to be from a subadult with most epiphyses separate from their respective long bone shafts. The status of epiphyseal union was unexpected because it indicated an individual much younger than the age known for the decedent, and the overall pattern of epiphyseal fusion was not consistent within the skeleton. The innominates were particularly immature compared with other skeletal elements. Fusion of the ischio-pubic ramus on both the left (Fig. 2) and right (Fig. 3) innominates is not complete. The ischopubic ramus is usually fused before puberty around age 7-10 in males (2,3). In addition, the ilium, ischium, and pubis are incompletely fused, and the ossifying triradiate cartilage is visible in the left innominate (Fig. 2). The appearance of the triradiate and ossification of the triradiate cartilage begins around age 9-10 years and fusion of the acetabulum follows around age 14-17 years in males (4). Nevertheless, the triradiate of this individual is still apparent and the acetabulum is incompletely formed (Fig. 3).

The epiphyseal fusion of other skeletal elements, such as the distal humerus (Fig. 4) suggests a slightly older individual. The trochlea is completely fused and the medial epicondyle is fused, with a visible line. Stevenson (5) gives an age range of 16–19 years and Ubelaker (3) an age range of 15–18 for the fusion of medial epicondyle in males. Fusion of the trochlea is estimated to be around ages 11–15 (3) and ages 12–17 for males (4).



FIG. 2—Left innominate with visible ossified triradiate cartilage and incompletely fused ischio-pubic ramus.



FIG. 3—Right innominate with incompletely fused acetabulum and incompletely fused ischio-pubic ramus.



FIG. 4-Right humerus.

Other synchondroses remain completely unfused and open, such as the spheno-occipital synchondrosis, proximal and distal femur, proximal humerus, distal radius and ulna, and the annular epiphyses of the verterbral centra all indicating an age of less than 18 years (2). The medial clavicle showed no signs of fusion and the ribs were underdeveloped for rib phase aging.

The degree of epiphyseal fusion seen in the skeleton indicated an individual much younger than 21–23 years. While few clinical studies measure epiphyseal fusion of the long bones directly, measures of skeletal maturity using hand and wrist radiographs indicate the timing and development of ossification centers are delayed and irregular in individuals with CP (6–8). Disrupted embryonic development, damage to the central nervous system, and brain injury are possible causes of delayed and/or uneven skeletal maturation and epiphyseal fusion in individuals with CP (6,9). Growth hormone deficiency and growth failure are possible complications stemming from neurochemical abnormalities associated with CP and can affect long bone length and skeletal maturation (10).

Malnutrition and disorders of feeding and swallowing are frequent in the CP population and are the most prevailing cause of growth and development retardation in children with severe CP (11–13). Henderson et al. (7) found in children with CP that large delays in skeletal maturation, including diminished linear bone growth, and low bone density were most prevalent in malnourished children. Medical records for this individual indicate that he was wheelchair bound and had difficulty swallowing and eating, a complication of CP that often puts individuals at risk for undernutrition and malnutrition (11). It is likely that malnutrition and neurological damage related to CP contributed to the delayed epiphyseal fusion seen in the skeleton, and the small size of the skeleton. Low bone density is often found in wheelchair-bound patients, and may have contributed to the light and fragile bones seen in this individual (14).

Environmental stressors such as undernourishment or malnutrition have a lesser affect on dental development than on skeletal growth and maturation (15,16). In contrast to the degree of epiphyseal fusion, dental development age estimates are somewhat closer to the chronological age of this individual. Radiographic and gross examination of the dentition revealed all permanent teeth except maxillary and mandibular third molars to be erupted and in occlusion with completely formed roots. Third mandibular molar roots were $\frac{3}{4}$ formed with apices open indicating a minimum age of 13 with a range of 13-20 years for males (17). Smith (16), reworking Moorrees et al.'s data (17) in order to produce a better age estimate for a given dental development stage, assigns an average age for similar root development of the mandibular third molars as 16.4 years for males. The dental age estimates are more consistent with the chronological age of the decedent than the skeletal age estimates for this individual. Table 1 summarizes the skeletal and dental age estimates for this individual.

TABLE 1—Summary of skeletal and dental age estimates.

| Element (and Degree of Fusion) | Age Estimate for Males | References |
|---|---------------------------|------------|
| Ischio-pubic ramus (unfused) | >10 years | (2) & (3) |
| Acetabulum and triradiate (triradiate present, acetabulum not completely fused) | >14 years | (4) |
| Medial epicondyle of humerus (fused with line) | 15-19 years | (5) & (3) |
| Trochlea of humerus (fused) | 11-15 years | (3) |
| | 12-17 years | (4) |
| Dentition | <13 years | (17) |
| | 16.4 years (average) | (16) |

Skeletal Analysis: Dental Pathologies

In addition to the skeletal and dental aging discrepancies, there were a number of dental pathologies. Figure 5 shows the overall occlusal surface of the maxillary teeth where a retained deciduous canine is visible on the right side, displacing the permanent canine. The right second premolar is also displaced and slightly rotated due to the roots of a retained deciduous premolar. Figure 6 shows a close-up of the occlusal surface of the left maxillary premolars and molars. The calculus buildup is extreme and some fiber or string has been incorporated into the calculus. Figure 7 shows a close-up of the anterior teeth of the mandible covered with extreme deposits of calculus. Despite the extensive calculus deposits, there were no caries or abscesses. Dental wear was extreme, suggesting chronic tooth grinding (bruxism), and is especially noticeable on the right mandibular molars (Fig. 8) where the cusps have been completely worn and large

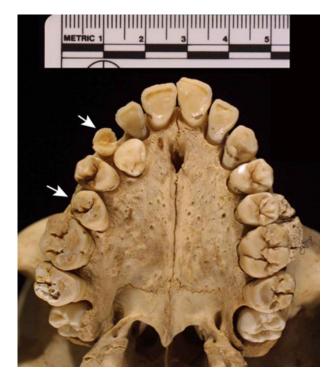


FIG. 5—Occlusal view of maxillae, top arrow points to retained deciduous canine, bottom arrow points to rotated second premolar.



FIG. 6—Left maxillary premolars and molars, note calculus and worn cusps.



FIG. 7—Anterior mandibular teeth, note calculus and visible dentin.



FIG. 8-Right mandibular molars with excessive wear.

areas of dentin are visible. Dental wear is also visible in Fig. 7 on the anterior mandibular teeth, and in Fig. 6 where the cusps of both the molars and premolars have been significantly worn.

Parafunctional oral habits are often present in individuals with CP and include bruxism, pacifier and finger sucking, and biting objects, all of which can impact the dentition and oral hygiene (18). Bruxism is found in significantly higher percentages of individuals with CP compared with non-CP controls (19). Plaque indexes are also significantly higher in children with CP compared with controls (19). The extreme dental wear and calculus deposits suggest significant parafunctional oral habits in this individual. Medical records indicate this individual was uncooperative for dental care, and the extent of calculus build-up and dental wear indicate that these conditions were likely untreated.

Discussion

Skeletal bone age delay and dental pathologies, such as those described in this individual, are common clinical findings in the CP population. This unique case demonstrates how numerous symptoms and complications of CP can manifest in one individual. Forensic cases documenting CP in the literature are scarce, so there are few cases with which to compare this analysis. However, Rosen et al.'s (20) analysis of how complications of CP may be significant to determining the manner of death is an important contribution to the forensic literature on this topic.

Unexpected observations that could not be corroborated with the CP literature include the underdeveloped innominates compared with the other skeletal elements. The unfused ischio-pubic ramus is inconsistent with the rest of the pattern of epiphyseal fusion seen in the skeleton and appears markedly more underdeveloped than other

skeletal elements. Bone age estimates for the rest of the skeleton (not including the underdeveloped ischio-pubic ramus) ranged between 11 and 19 years. These skeletal age estimates are consistent with some bone age delay and delayed skeletal maturation routinely seen in CP patients. This individual experienced significant disability related to CP and most likely represents a more extreme example of delayed skeletal maturation and growth. Much of the bone age delay, delayed epiphyseal fusion, and small bone size likely stem from neurological damage related to CP, difficulties eating and swallowing, and consequent undernutrition and malnutrition.

The dental wear and calculus deposits in this case are extreme, even for CP cases documented in the clinical literature. This suggests that significant parafunctional oral habits went untreated for some time, and medical documents indicate that this individual was uncooperative during dental appointments.

This analysis represents one unique forensic case with CP that will add to the scarce literature examining the affect of CP and other related disabilities on skeletal age and dentition. Perhaps the most striking and significant feature associated with CP is dismaturation. In cases where there is likelihood that remains are those of a person who had CP, forensic anthropologists need to be aware of the probability that standard developmental indicators may lead to an underestimation of age at death. In cases where there is no information about a health condition during life that may have affected development, this article may alert investigators to some of the osteological signs of CP and the probability that age indicators may be misleading. By documenting the effects of CP in this case, we hope to provide a framework for future cases where complications from disabilities play a role in interpreting the biological profile.

Acknowledgments

We thank Dr. Todd Fenton (Michigan State University) and Kyle Hoskins (Michigan State Police) for their assistance with the analysis and recovery of this case, and Jamie Minns (MSU Forensic Anthropology Lab) for her help with some of the images.

References

- Bax M, Goldstein M, Rosenbaum P, Leviton A, Paneth N, Dan B, et al. Proposed definition and classification of cerebral palsy, April 2005. Dev Med Child Neurol 2005;47:571–6.
- Krogman W, Iscan MY, editors. The human skeleton in forensic medicine. Springfield: Charles C. Thomas, 1986.
- 3. Ubelaker DH. Human skeletal remains, 2nd ed. Washington, DC: Taraxacum, 1989.
- 4. Scheuer L, Black S. The juvenile skeleton. London: Elsevier, 2004.
- Stevenson PH. Age order of epiphyseal union in man. Am J Phys Anthropol 1924;7:53–93.
- Ihkkan DY, Yalcin E. Changes in skeletal maturation and mineralization in children with cerebral palsy and evaluation of related factors. J Child Neurol 2001;16:425–30.
- Henderson RC, Gilbert SR, Clement ME, Abbas A, Worley G, Stevenson RD. Altered skeletal maturation in moderate to severe cerebral palsy. Dev Med Child Neurol 2005;47:229–36.
- Gilbert SR, Gilbert AC, Henderson RC. Skeletal maturation in children with quadriplegic cerebral palsy. J Pediatr Orthop 2004;24:292–7.
- Kong CK, Tse PWT, Lee WY. Bone age and linear skeletal growth of children with cerebral palsy. Dev Med Child Neurol 1999;41:758– 65.
- Coniglio SJ, Stevenson RD, Rogol AD. Apparent growth hormone deficiency in children with cerebral palsy. Dev Med Child Neurol 1996;38:797–804.
- Rogers B. Feeding method and health outcomes of children with cerebral palsy. J Pediatr 2004;145(2 Suppl):S28–32.

274 JOURNAL OF FORENSIC SCIENCES

- Kong CK, Wong HS. Weight-for-height values and limb anthropometric composition of tube-fed children with quadriplegic cerebral palsy. Pediatrics 2005;116:839–45.
- Stallings VA, Charney EB, Davies JC, Cronk CE. Nutrition-related growth failure of children with quadriplegic cerebral palsy. Dev Med Child Neurol 1993;35:126–38.
- Pluskiewicz W, Drozdzowska B, Lyssek-Boro A, Bielecki T, Adamczyk P, Sawaryn P, et al. Densitometric and quantitative ultrasound measurements and laboratory investigations in wheelchair-bound patients. J Clin Densitom 2006;9:78–83.
- 15. Ubelaker DH. Estimating age at death from immature human skeletons: an overview. J Forensic Sci 1987;32:1254-63.
- Smith BH. Standards of human tooth formation and dental age assessment. In: Kelley MA, Larsen CS, editors. Advances in dental anthropology. New York: Wiley-Liss Inc., 1991;143–68.
- 17. Moorrees CF, Fanning EA, Hunt EE. Age variation of formation stages for ten permanent teeth. J Dent Res 1963;42:1490–502.

- Ortega AOL, Guimaraes AS, Ciamponi AL, Marie SKN. Frequency of parafunctional oral habits in patients with cerebral palsy. J Oral Rehabil 2007;34:323–8.
- Dos Santos MTBR, Masiero D, Novo NF, Simionato MRL. Oral conditions in children with cerebral palsy. J Dent Child 2003;70:40–6.
- Rosen RS, Armbrustmacher V, Sampson BA. Mortality in cerebral palsy (CP): the importance of the cause of CP on manner of death. J Forensic Sci 2003;48:1144–7.

Additional information and reprint requests: Mary S. Megyesi, M.S. Department of Anthropology Michigan State University 354 Baker Hall East Lansing, MI 48824 E-mail: megyesim@msu.edu