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Multiple supernumerary teeth in a likely syndromic individual from prehistoric Illinois

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ABSTRACT

Objective: This paper reports the first published case of a prehistoric human with five or more supernumerary teeth. Such cases are often neglected in paleopathology, in part due to a gap between the medicodental and anthropological literature leading to the view in anthropology that supernumerary teeth are curious anomalies rather than pathologies.

Design: Reconstruction and pathological description of the skeletal remains were performed according to standard osteological protocols. Each supernumerary tooth was categorized based on its morphology, location, and orientation. The dental characteristics of the individual were compared to published norms for incidences of syndromic and non-syndromic supernumerary teeth and a differential diagnosis was subsequently performed.

Results: Six supernumerary teeth and one deciduous tooth were identified. Additionally, the individual suffered from impacted teeth, dilacerated roots, and extensive sutural anomalies (including retention of the metopic suture into adulthood and an unusually high number of sutural bones). The morphology and location of the supernumerary teeth, in conjunction with the suite of other symptoms, are highly unusual among non-syndromic patients and therefore are indicative of a complex genetic disorder.

Conclusions: The individual reported here almost certainly suffered from a genetic disorder or syndrome resulting in extensive dental and sutural abnormalities. Despite a lack of post-cranial involvement, a tentative diagnosis of Cleidocranial Dysplasia was made on the basis that mutations in the *RUNX2* gene may cause the dental symptoms without any pathology of the clavicle.

1. Introduction

Supernumerary teeth (teeth in addition to the usual 20 deciduous or 32 permanent, whether impacted or erupted) are one of the most common developmental anomalies in humans (Klein et al., 2013). They are associated with retention of deciduous teeth, displacement or rotation of adjacent teeth, crowding, root dilacerations (angulation or curvature), or impaction (Liu & Chen, 2014) and therefore are of clinical importance. Prevalence varies by population from 0.1% to 3.8% (Bereket, Çakir-Özkan, Sener, Bulut, & Bastan, 2015) with an average global frequency of 2.6% (Chou et al., 2015). The cause of supernumerary teeth may be due to retention and reactivation of dental lamina fragments, proliferation of the dental lamina due to defective epithelium-mesenchyme signaling, or division of a single tooth bud (D'Souza & Klein, 2007). Although the definitive etiology remains unknown, the dental lamina hyperactivity hypothesis has the most support (Takahashi et al., 2016). The familial incidence of supernumerary teeth indicates a genetic basis but it does not follow simple Mendelian inheritance and is poorly understood (Orhan, Ozer, & Orhan, 2006).

The majority of cases are idiopathic but supernumerary teeth are also associated with a number of genetic syndromes, notably Cleidocranial Dysplasia and Familial Adenomatous Polyposis (Lubinsky & Kantaputra, 2016). Syndromic cases of supernumerary teeth generally differ from non-syndromic cases in the number and position of extra teeth. Over 70% of non-syndromic cases of supernumerary teeth involve only a single extra tooth (Bereket et al., 2015; Rajab & Hamdan, 2002), most commonly positioned between the upper central incisors (Bozkurt, Bezgin, Tüzüner Öncül, Göçer, & Sarı, 2015). Only 0.049% of cases of multiple supernumerary teeth, defined in dentistry as five or more supernumerary teeth in a single patient, are not associated with complex syndromes (Yagüe-García, Berini-Aytés, & Gay-Escoda, 2009). Syndromic multiple supernumerary teeth generally show bilateral symmetry and exhibit unusual morphology (Açikgöz, Açikgöz, Tunga, & Otan, 2006; Yassin & Hamori, 2009). Such cases are also frequently accompanied by cranial suture abnormalities due to their shared signaling pathways during cytodifferentiation (De Coster, Mortier, Marks, & Martens, 2007). Regardless of syndrome status, all supernumerary teeth are significantly more common in males than in females and in

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the maxilla than in the mandible (Jung, Kim, & Cho, 2016; Yagüe-García et al., 2009).

This paper describes an individual from prehistoric Illinois with six supernumerary teeth and a suite of sutural abnormalities. To the author's knowledge, this publication represents the first paleopathological description of an individual with multiple supernumerary teeth. Despite their overall prevalence in modern human populations, supernumerary teeth are rarely reported in the paleopathological literature. This phenomenon is in part due to the prevailing characterization of supernumerary teeth as a normal variant, thereby ignoring their clinical relevance. Standard visual and textual recording methods in human osteology take into account the location of the tooth within the jaw but do not easily allow description of the morphology of the supernumerary tooth or the common oral side-effects of supernumerary teeth (Buikstra & Ubelaker, 1994). Even in the most in-depth paleopathology reference books, supernumerary teeth are described in terms of possible locations but not in any pathological context (Ortner, 2003) or as isolated symptoms of specific syndromes but not discussed under dental pathology (Aufderheide & Rodriguez-Martin, 2008). This paper attempts to bridge the gap between the medicodental literature, which focuses on soft-tissue diagnosis in living patients and symptom management, and paleopathology by presenting an in-depth description and differential diagnosis of a likely syndromic prehistoric individual with six supernumerary teeth.

2. Materials and methods

The individual described here (KM˚Kn1-04) was encountered during a systematic study of dental wear at Koster Mounds, a skeletal series of uncertain cultural affiliation and chronology from prehistoric Illinois. The site was excavated by Gregory Perino of the Gilcrease Institute in the early 1960s. KM˚Kn1-04 was buried in a flexed position without any mortuary goods or artifacts in a grave eight inches below the surface of a natural knoll overlooking the Illinois River Valley (Perino, 1973). Nine carbon dates from the skeletal series at Koster Mounds have been obtained, all of which place it roughly within the Late Woodland period of Midwest prehistory from 500 to 1000 CE (Berger & Protsch, 1989; Conner, 1984; Crane & Griffin, 1966). However, substantial evidence points to an earlier use of the site and its exact age is currently under re-evaluation by the author.

The skeletal remains for KM˚Kn1-04 are fairly fragmentary. The skull was reconstructed with Duco Cement (an acetone-soluble adhesive) using thin wooden reinforcements to visualize the greatest possible extent of pathology (Fig. 1). Every effort was made to preserve the original morphology and minimize distortions. Although the postcranial remains were well-represented, they did not include any complete long bones (Fig. 2). Reconstruction was attempted for postcranial elements but proved minimally productive.

The individual was identified as male based on robust supraorbital ridges and a narrow greater sciatic notch using standard protocols for skeletal analysis (Buikstra & Ubelaker, 1994). All sexually dimorphic features of the skull aside from the supraorbital ridges (including the zygomatic bones, mastoid processes, nuchal crest, and mental eminence) were either incomplete or affected by pathology (Fig. 1). The fragmentary nature of the postcranial elements precluded the use of metric sexing techniques, which are generally regarded as the most reliable method of sex assessment (Spradley & Jantz, 2011), or of any pelvic morphology aside from the greater sciatic notch (Fig. 2). Although supraorbital ridges are not the most reliable cranial feature for sex assessment and cannot be safely used in isolation (Walker, 2008), the similarly masculine morphology of the greater sciatic notch lends credence to the male sex assessment based on cranial morphology (Garvin, 2012). Age assessment was similarly affected by poor preservation and pathology. The individual was tentatively identified as being 21–24 years old at his time of death based on an unfused sternal

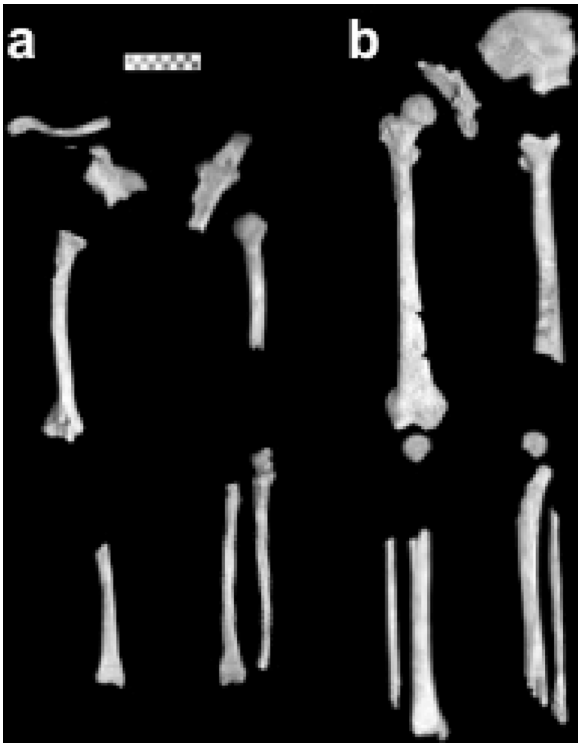

Fig. 2. The postcranial remains from the upper limb (a) and lower limb (b) associated with the individual. No complete long bones were recovered (the posterior aspect of the right femur is missing and prevented its measurement). The left greater sciatic notch is quite narrow. Additional remains not shown include four rib fragments; two broken cervical vertebrae; all right carpals except for the pisiform; the left scaphoid, pisiform, and hamate; nine hand phalanges; the right calcaneus; the right and left talus; and the right cuboid.

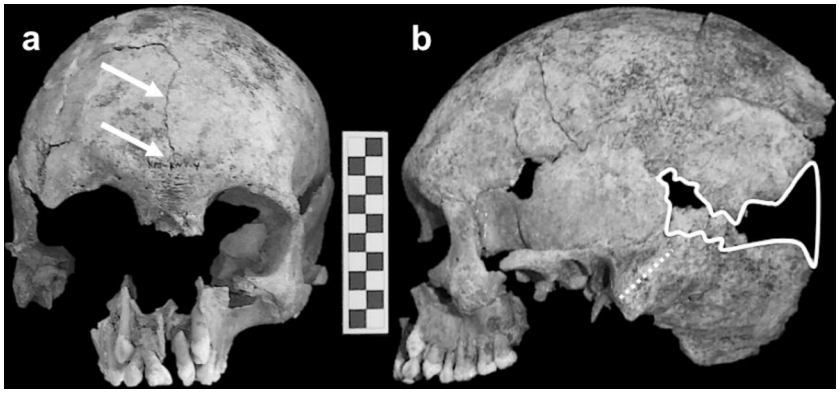

Fig. 1. Anterior (a) and left lateral (b) images of the cranium. Fragments were reconstructed by hand with every attempt made to preserve the original morphology. The metopic suture is retained into adulthood (white arrows) both ecto- and endocranially. The majority of the occiput of the skull is composed of a large number of sutural bones (area outlined by the white line). There is a depression anterior to the mastoid process (white dotted line) indicative of delayed or anomalous closure of the suture between the petromastoid and squamous portions of the temporal bone. The gap between the zygomatic and temporal bones in the zygomatic arch is the result of developmental hypoplasia rather than taphonomy based on the rounded edges of the temporal and zygomatic arches and the lack of any visible cancellous bone.