Brief Communication: Unusual Finding at Pueblo Bonito: Multiple Cases of Hyperostosis Frontalis Interna

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ABSTRACT Hyperostosis frontalis interna (HFI) is a disease characterized by excess bone growth on the internal lamina of the frontal bone and, occasionally, other cranial bones. Although the disease is fairly common in modern populations, its etiology is poorly understood. Hyperostosis frontalis interna has been identified in antiquity, primarily in the Old World, but with a much lower frequency than in modern groups. The purpose of the present study is to report multiple cases of HFI at Pueblo Bonito (Chaco Canyon, New Mexico). Twelve out of 37 adults with observable frontal bones exhibited HFI, ranging from mild to severe, including 11 females and one male. This is the first published case report of HFI in archaeological remains from the New World having a frequency comparable with modern groups. Most archaeological cases of HFI are isolated, so comparative data for multiple cases at one site are rare. The results of this study emphasize the importance of looking for HFI in archaeological remains, although it is rarely observed. Possible genetic and environmental factors for the high frequency of HFI at Chaco Canyon are considered, but additional research is needed to discover the etiology and to better understand why HFI sometimes occurs at modern frequencies in ancient populations. Am J Phys Anthropol 130:480–484, 2006.

Hyperostosis frontalis interna (HFI), originally described by Morgagni in the 18th century, is characterized by irregular bony nodules on the inner table of the frontal bone. In advanced cases, the abnormal bone may extend onto the parietals and the occipital. The bony accretion is nonspecific and generally benign, but severe HFI is sometimes associated with mental disturbance, e.g., when it occurs as a symptom of Stewart-Morel syndrome (Phillips, 1997). Perou (1964) suggested a number of possible causative factors for HFI, including heredity, endocrine disorders, dysplasia, dystrophy, neoplasia, and trauma. Despite numerous studies on the subject, the etiology of HFI is still uncertain (She and Szakacs, 2004).

Hyperostosis frontalis interna is a common finding in clinical settings, but there is considerable variation in the reported incidence of HFI in modern populations. This condition is most prevalent in postmenopausal females, with frequencies of 40–62% reported (Resnick, 2002). In contrast, HFI is rarely reported in an archaeological context. Most reports of HFI in archaeological remains are isolated cases (e.g., Armelagos and Chrisman, 1988; Anderson, 1993; Rühli and Henneberg, 2004), but Lazer (1996) reported observing HFI in 43 out of 360 skulls (11.9%) from Pompeii. Hershkovitz et al. (1999) surveyed a large sample of historic and archeological skeletons (n = 2,019) from the Smithsonian Institution for HFI, including 1,012 Native American skeletons representing Alaska, Arkansas, California, Illinois, Louisiana, Mississippi, New York, Ohio, Pennsylvania, South Dakota, and Virginia. The sites included remains dating from the 16th–17th centuries AD. No cases of HFI were found in this large sample, providing further support for the rarity of HFI in antiquity.

The importance of systematic analysis of Native American remains currently housed in museum collections is highlighted by the recent discovery of 12 cases of HFI in skeletal remains from Pueblo Bonito, New Mexico (AD 860–1150), during standard documentation by the Repatriation Osteology Laboratory (ROL) at the National Museum of Natural History (NMNH), Smithsonian Institution. While the survey for HFI by Hershkovitz et al. (1999) in the NMNH collections was extensive, the ROL completed an analysis of over 4,000 Native American individuals since 1998. Such large data collections are particularly important for identifying relatively rare conditions. This paper describes individual cases of HFI from Pueblo Bonito, and discusses the findings in the context of documented frequencies in modern, historic, and prehistoric populations.

MATERIALS AND METHODS

Pueblo Bonito is the largest Anasazi great house in the Chaco Canyon Complex of the San Juan Basin, New Mexico. The 651 rooms, reaching up to four stories tall, were constructed in several stages. As construction proceeded, portions of the Pueblo were abandoned, so all rooms were never contemporaneously occupied. Through a comprehensive mortuary analysis and craniometric study of a broader series of Chaco Canyon interments, Akins (2003) concluded that Pueblo Bonito was a place of high-status residence and interments, with status ascribed rather than achieved. She also maintained that burial clusters likely represent kinship groups.

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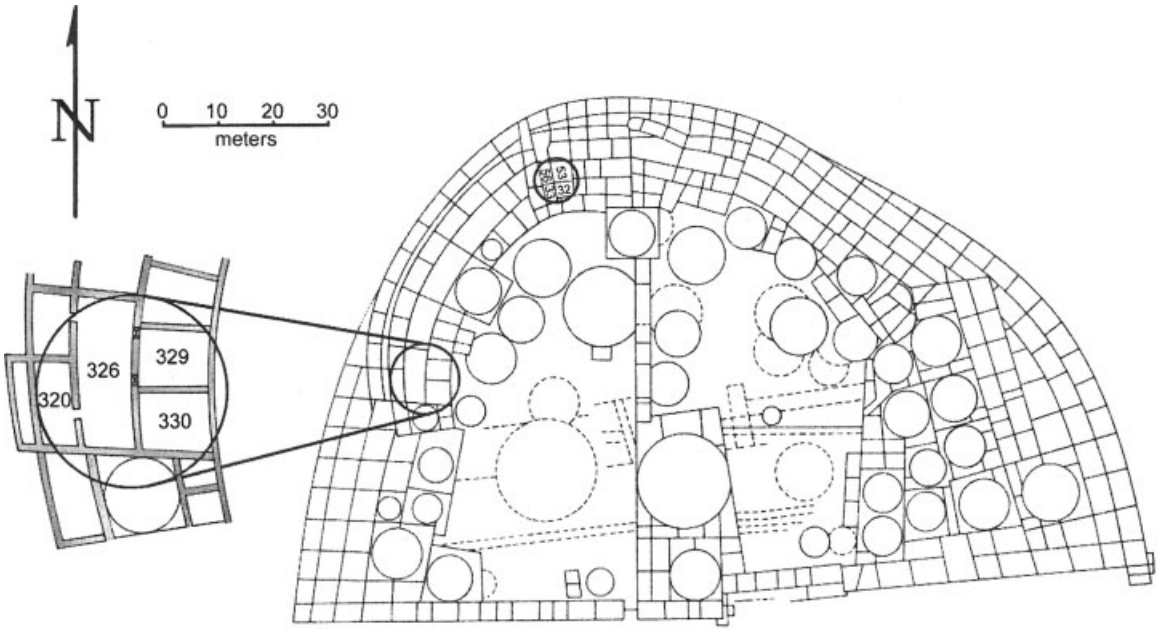

Fig. 1. Floor plan of Pueblo Bonito, with circled areas outlining room clusters with multiple burials. Western cluster is enlarged to show rooms containing remains analyzed in this study, while Northern cluster is only outlined (adapted from Akins, 2003).

The excavation of Chaco Canyon burials has a long and often poor history of documentation (Akins, 1986). Human interments are scattered throughout Pueblo Bonito, but the majority of the 131 known burials were discovered in two clusters, each involving four adjacent rooms with no outside access (Fig. 1, Table 1). These rooms were located in the "Old Bonito" section of the site, with a construction date from AD 860–935. However, ceramics associated with the burials indicate a later interment, between roughly AD 1020–1150 (Akins, 1986, 2003). The Northern cluster was excavated by the American Museum of Natural History/Hyde Expedition of 1896–1899, and the Western cluster by the National Geographic Society Expedition of 1920–1927.

Our analysis included all individuals currently curated at the NMNH. All remains were excavated from the Western cluster. Only adults with a minimum age of 18 years with complete or nearly complete frontal bones were included in computing HFI frequencies, resulting in a sample of 12 males and 25 females. Age and sex were determined using standard osteological indicators (Buikstra and Ubelaker, 1994), and are reported in Table 1. In both cases, greater weight was assigned to pelvic indicators over cranial morphology. In four cases with HFI (327065, 327118, 327125, and 327116), only crania were available. Standard protocol in the ROL includes identification of pathological changes through gross morphology and analysis of three standard cranial radiographs (lateral, frontal, and basilar; Ousley et al., 2005). The endocranial surface of the skull is viewed with a flexible penlight. Although mild cases of HFI are not always visible on radiographs (Hershkovitz et al., 1999) and it is not always possible to observe the entire endocranial surface of the frontal bone, most cases of HFI should be identified when following the standard protocol of the ROL.

The severity of HFI was scored using the four types defined by Hershkovitz et al. (1999). Briefly, these types are defined as follows. Type A includes cases with small, discrete nodules generally smaller than 10 mm in diameter. Type B includes cases with nodular bony overgrowth without discrete margins, affecting less than 25% of the frontal endocranial surface. Type C is more extensive nodular bony overgrowth, affecting up to 50% of the frontal endocranial surface. Type D is characterized by continuous bony overgrowth affecting more than 50% of the endocranial frontal bone. Hershkovitz et al. (1999) elevated the classification to the next level if other bones, such as the parietal, were involved. In the present study, involvement of the parietal bone is indicated separately, so the morphology of the frontal bone follows the type description.

TABLE 1. Sample age and sex distribution and HFI scores

SI number	Room	Sex	Age	HFI score	Parietal involved
327065	320A	F	20–35	B	No
327059	320A	F	40–50	C	Yes
327066	326	F	35–45	B	No
327067	326	F	35–50	B	No
327070	326	F	40–45	B	No
327071	326	F	35–45	B	No
327075	326	F	45–50	B	No
327076	326	F	40–55	C	Yes
327084	330	M	35–45	A	No
327116	329	F	45+	C	Yes
327118	329	F	35–45	B	No
327125	329	F	40–60	A	No

RESULTS

Hyperostosis frontalis interna was identified in 12 out of 37 adults (32.4%) with observable frontal bones, including 1 out of 12 males (8.3%) and 11 out of 25 females (44.0%). All individuals but one were over age 35 years (Table 1). As shown in Table 1, the sample from Pueblo Bonito included two individuals with type A HFI, seven with type B, and three with type C. All three individuals with type C HFI also exhibited involvement of the parietal bone. With the

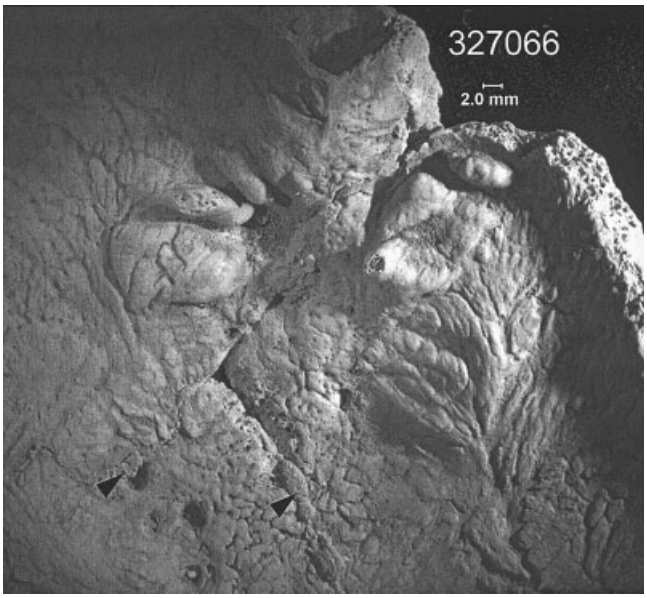

Fig. 2. Endocranial image of SI 327066, a 35–45-year-old female at HFI stage B. Frontal crest is oriented vertically, and is obscured by expansive bony nodules. Arrowheads indicate glue used in past reconstructive efforts, surrounded by possible hemorrhagic response.

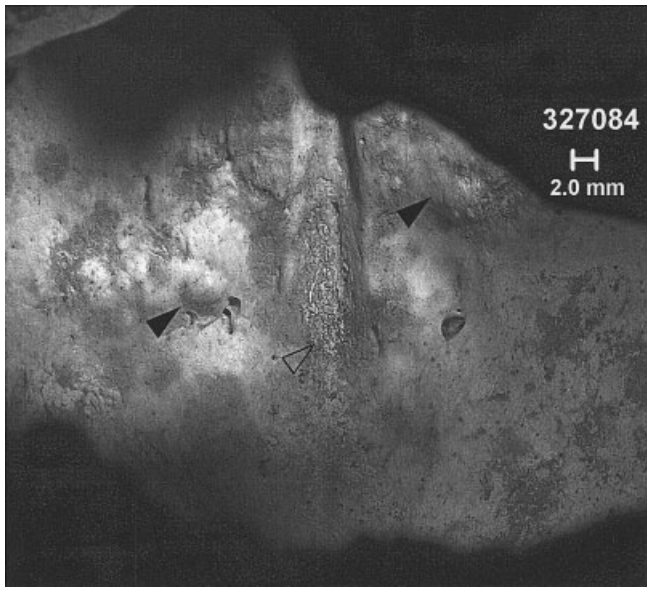

Fig. 4. Endocranial image of SI 327084, a 35–45-year-old male at HFI stage A. Frontal crest is oriented vertically with bony nodules on either side, as indicated by solid arrowheads. Open arrowhead pinpoints inflammatory or hemorrhagic response.

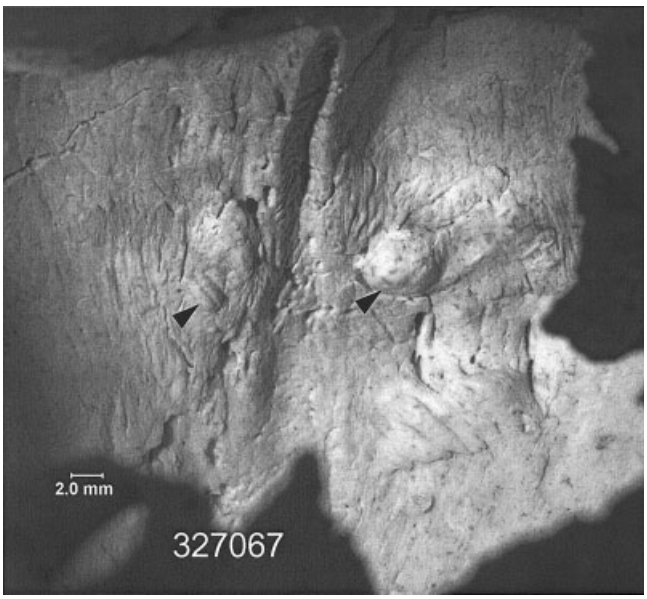

Fig. 3. Endocranial image of SI 327067, a 35–50-year-old female at HFI stage B. Frontal crest is oriented vertically with bony nodules on either side, as indicated by arrowheads.

exception of one mild case (327125), all cases show bilateral involvement and the characteristic "knobby" appearance of HFI lesions. The midline is unaffected all cases but one. No evidence of periostitis or woven bone is present. In addition to HFI, one individual (327066) exhibits additional lesions that reflect other disease processes, including ectocranial lesions on the frontal and parietals, consistent with treponemal disease and the presence of a possible hemorrhagic response with associated porosity and vascular channels on the endocranial surface, near the HFI lesions. The remaining individuals do not exhibit endocranial vault lesions other than those attributable to HFI. Figures 2–4 give examples of the observed lesions from three individuals, including two females and one male. Figure 3 illustrates the HFI and possible hemorrhagic lesions seen in 327066.

All cases of HFI were first identified by gross examination of the endocranial surface. Subsequent examination of the radiographs showed observable HFI for all type C cases, while types A and B were not always visible on the standard views. A search of ROL databases showed no other identified cases in 4,552 Native American frontal bones from sites throughout the United States. This total includes 130 adults with observable frontal bones from Hawikku, 11 from Guisewa, and 42 from Jemez, all from New Mexico.

DISCUSSION

Hershkovitz et al. (1999) and Anton (1997) discussed the differential diagnosis of HFI from other sources of cranial hypertrophy, including tumors, pregnancy osteophytes, Frölich syndrome, Paget's disease, acromegaly, leontiasis ossea, and fibrous dysplasia. In HFI, the ectocranial surface and midline are usually unaffected, and lesions are often bilateral. HFI can be differentiated from other diseases based on the morphology and distribution of lesions. Osteomas are generally ectocranial and are usually not bilateral. Frölich syndrome and Paget's disease generally involve the cranial base. Acromegaly, Paget's disease, and fibrous dysplasia both affect the ectocranium and diploic space. Pregnancy osteophytes are more prevalent on the ectocranial vault than on the endocranial surface, and they predilect the venous sinuses. None of these disease processes are consistent with the observed lesions at Pueblo Bonito.

The presence of HFI at Pueblo Bonito is particularly interesting, given the absence of HFI in the North American archaeological record. There may be some overlap in the Native American skeletal sample studied by Hershkovitz

et al. (1999) and the 4,552 remains documented by the ROL, but the combined sample represents most of the United States. The skeletal remains documented by the ROL include a larger Southwest sample, which is important for comparisons with the Chaco Canyon sample. Akins (1986, p. 49) also analyzed 135 individuals recovered from small sites within Chaco Canyon, evaluating them for cranial deformation, pacchonian pits, and problematic subadult lesions described as "endocranial striae and pitting." It may thus be inferred that such examination would have uncovered HFI if it was present in the small-site Chaco burials.

In their literature review and survey of historic and archaeological samples, Hershkovitz et al. (1999) found that the incidence of HFI increased during the 20th century among females, and remained constant or increased slightly for males. They found HFI in 25% of females and 5% of males in a large skeletal sample. (n = 1,007) from the early 20th century. Overall, their findings were consistent with modern clinical observations, with older females exhibiting the most frequent and severe HFI. While HFI was even identified in fossil hominids such as Sangiran, Gibraltar 1, and Shanidar 5 (Anton, 1997), HFI is apparently rare in antiquity, with isolated cases identified at various sites. However, the present study and that by Lazer (1996) on Pompeii suggest that this disease can occur at higher frequencies in ancient populations. Lazer (1996) concluded that the presence of HFI at Pompeii showed that longevity was comparable to modern groups, but estimated ages and sexes were not reported for individuals with HFI, apparently because the sample was commingled.

The present study includes the first reported cases of HFI in archaeological remains from the New World with a frequency similar to that seen in modern groups. An abstract by Everett (2002) reported on the only other example of HFI in Native American skeletal remains. Specifically, the presence of HFI in three individuals over 50 years of age (2 female and 1 male) from the Pete Klunk Middle and Late Woodland Mounds site in Illinois was reported, but there are no published photographs. All three cases were reported as mild, so at present, Pueblo Bonito is the only Native American sample with significant pathognomic pathological changes associated with HFI.

Historic cases of HFI are documented in the US, including a single case at the First African Baptist Church of Philadelphia (1823–1841) (Angel, 1987). Phillips (1997) reported a total of 10 out of 18 females (including pre- and postmenopausal women) and 16 out of 27 males with HFI associated with the Oneida County Almshouse in New York state, which operated during the 19th century. Although this is an example of a site with a high frequency of HFI, it represents a societal subset of the population that may overrepresent the frequency in the general population.

Rühli et al. (2004) determined that HFI was more frequent in males in antiquity, based on previously reported isolated cases in the Old World. The present study does not provide support for the idea that HFI was more frequent in males in antiquity, and is more consistent with the pattern observed in modern populations, with HFI affecting females much more frequently than males. These seemingly contradictory patterns are not surprising, given that HFI is a generalized pathological condition with an unknown etiology and variable clinical association.

Although it is not possible to isolate the cause of HFI at Pueblo Bonito, several contributing factors can be considered. An unknown genetic predisposition or common environmental exposure could account in part for the high frequency of HFI within the Western burial cluster, as a mortuary and craniometric study concluded that these burials represent higher-status individuals from "closely related groups," involved in a "family burial facility" (Akins, 2003, p. 101, 105). Armelagos and Chrisman (1988) proposed that HFI was rare in antiquity because the life span was shorter. Other researchers further suggested that increased longevity has resulted in changes in the human life cycle and hormone levels, indirectly causing higher frequencies of HFI in modern populations (Hershkovitz et al., 1999; Rühli and Henneberg, 2002; Rühli et al., 2004). If this is the case, the Pueblo Bonito sample may represent a population with a female life cycle similar to that in modern groups, including a long duration between menarche and menopause, as well as less time spent pregnant and nursing than in contemporaneous groups. Associations of HFI with obesity, virilism, and diabetes as clinical diagnostic features in several syndromic conditions also spurred hypotheses of endocrine imbalances in HFI development, including specific links to levels of leptin, estrogen, androgens, prolactin, and progesterone (Hershkovitz et al., 1999; Rühli et al., 2004).

The effect of dietary phytoestrogens on human health has had an increasing presence in the clinical literature during the past decade. Phytoestrogens include isoflavones and lignans, found in various foods such as soy, linseed, grains, and vegetables. Although corn, which does not include these compounds, was the dietary staple at Chaco Canyon, a variety of other plants and herbs were consumed. Dietary phytoestrogens have biological effects on pre- and postmenopausal women, including alterations in estrogen metabolism and possible prevention of bone loss (Cassidy and Faughnan, 2000). However, the extent and complexity of the interactions between diet and biological function are still not well-understood.

Hyperostosis frontalis interna occurs much more commonly today than it did in ancient times. However, cases such as those reported here and the one reported by Lazer (1996) in skeletal remains from Pompeii suggest that it did occur with a high frequency in certain ancient populations. Isolating the cause of the disease in these cases is not possible at present, but additional cases may help shed light on patterns among affected groups. In addition, HFI may be underrepresented in ancient skeletal remains due to its location inside the cranial vault. These points highlight the importance of looking for HFI in ancient skeletal remains, even though the condition appears to be uncommon.

CONCLUSIONS

Hyperostosis frontalis interna was observed in 12 out of 37 individuals from Pueblo Bonito. The observed results are considered within the greater context of HFI in ancient populations in general, and in North America in particular.

1. Although HFI may be underreported in ancient populations, it is clearly rare in ancient populations from North America, based on a previous study by Hershkovitz et al. (1999) as well as the research conducted by the ROL at the Smithsonian Institution, which together represent a sample of between about 4,500–5,700 individuals.
2. The frequency of HFI at Pueblo Bonito is consistent with the frequency observed in modern populations.

3. The high frequeny of HFI at Pueblo Bonito may indicate that females had a life cycle similar to that in modern populations.
4. A genetic predisposition to HFI at Pueblo Bonito is possible; previous research suggests that the Western burial cluster represents closely related groups.
5. A common environmental factor, such as the presence of dietary phytoestrogens, could also affect the frequency of HFI, but the complexity of possible interactions is not well-understood.
6. Hyperostosis frontalis interna occasionally occurs at modern frequencies in ancient populations. Regardless of whether this is because it is underreported or because it is rare, additional reports are needed to better understand the etiology of the disease.

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