J Forensic Sci, Jan. 2005, Vol. 50, No. 1
Paper ID JFS2004173
Available online at: www.astm.org

William Devriendt,[1] *Ph.D.; Marie-Dominique Piercecchi-Marti,*[1,2] *M.D., Ph.D.; Pascal Adalian,*[1] *Ph.D.; Alain Sanvoisin,*[3] *M.D.; Olivier Dutour,*[1] *M.D., Ph.D.; and Georges Leonetti,*[1,2] *M.D., Ph.D.*

Hyperostosis Frontalis Interna: Forensic Issues

ABSTRACT: The use of pathological conditions in age and sex determination, important factors in personal identification, is not widespread in anthropology and legal medicine. Hyperostosis frontalis interna (HFI) is a bone condition that mainly affects the inner table of the frontal bone. Although there are numerous publications on the subject, at the present time its etiology remains obscure. Several associations of symptoms, whose incidence varies according to the population studied, have been described. Age and gender appear to be linked with the preponderance of this condition, as does the presence of behavioral disturbances. The aim of our study, based on a series of 1532 autopsies, was to define the incidence and the associations observed with other pathological conditions. Thirteen cases of HFI were identified (0.8% of autopsies), 12 women and one man whose mean age was 59.15 years (range: 42–79 years). All had behavioral disturbances and most were under psychiatric care. This study emphasizes the value of this condition in medico-legal identification.

KEYWORDS: forensic science, hyperostosis frontalis interna, medico-legal anthropology, identification

Identification in medico-legal anthropology is a recurrent problem. In order to make an estimation of the age and sex of an individual in a skeletal state, a variety of methods, both qualitative and quantitative, are available to the specialist. The various taphonomic factors that determine skeletal preservation sometimes exclude the use of certain anatomical parts for identification of the body. The observation of certain conditions whose incidence is related to the sex and age of the individuals is a rarely explored area of investigation. Among these conditions, hyperostosis frontalis interna (HFI) is a particularly significant example.

Hyperostosis frontalis interna is manifested by the accretion of bone on the inner table of the frontal bone. Bony overgrowths, all of similar size, are separated from each other by deep grooves and distributed symmetrically on each side of the *crista frontalis*, which is spared (Fig. 1). The extent of HFI is variable, leading to its classification in 4 stages.

The unknown etiology of HFI has incited numerous clinicians to examine the "symptoms" that are most frequently associated with this bony lesion. The result is a considerable volume of literature dealing with diverse accompanying symptoms whose frequency varies considerably from one publication to another. Nevertheless, the very marked female predominance is widely accepted, as is the association of three "symptoms" that are virilism, obesity and behavioral disturbances, known as Morgagni-Stewart-Morel syndrome.

The aim of this four-year prospective study was on the one hand to determine the incidence of this condition as revealed by medico-legal autopsies, and on the other hand to search for these "symptoms" that could be useful for the identification of an individual.

[1] UMR 6578-CNRS/Université de la Méditerranée, Faculté de Médecine de Marseille, 27 Boulevard Jean Moulin, 13385 Marseille Cedex 05, France.
[2] Laboratoire de Médecine Légale, Faculté de Médecine de Marseille, 27 Boulevard Jean Moulin, 13385 Marseille Cedex 05, France.
[3] Service de Médecine Légale, Faculté de Médecine de Marseille, 27 Boulevard Jean Moulin, 13385 Marseille Cedex 05, France.

Received 24 April 2004; and in revised form 16 July 2004; accepted 31 July 2004; published 15 Dec. 2004.

Material and Methods

Our study sample consisted of a series of 1532 autopsies carried out in the department of legal medicine of the Hôpital de la Timone, Marseille, France, between 1 January 1999 and 31 December 2002.

Hyperostosis frontalis interna results in mild to marked symmetric thickening of the inner table of the frontal bone, producing a wavy or "choppy sea" appearance. The midline and occipital areas are spared. The thickening generally does not go over the limits of the frontal bone but can extend to the upper orbital plate and to the parietal parts of the calvaria in extreme cases, even leading to partial brain atrophy. Whatever the degree of involvement, only the inner bony tables are modified.

In order to clearly distinguish the different levels of the condition, we used the classification proposed by Hershkovitz et al. (1) according to the extent of the lesion:

Type A: characterized by isolated or multiple bony nodules, unilateral or bilateral, often found on the anteromedial part of the frontal bone, and not exceeding 10 mm in size.

Type B: marked by more numerous bony overgrowths that may cause slight elevation of the inner table of the frontal bone.

Type C: at this stage, the bony overgrowths are more extensive, with irregular thickening of the frontal endocranial surface.

Type D: the frontal bone is very thick. Bony overgrowth involves more than 50% of the endocranial surface. The area has a rough appearance.

An anterior radiograph of the skull was obtained before the cranium was opened.

Results

Thirteen cases of HFI were observed, 12 women and one man. They were aged between 42 and 79 years (mean 59.15 years) (Table 1).

We observed only types C (38.5%) and D (61.5%). HFI led to thickening of the frontal bone, which measured from 15 to 28 mm (Fig. 2). In one case, HFI was associated with asymmetric cerebral atrophy, in two cases with pituitary hypertrophy and in one case

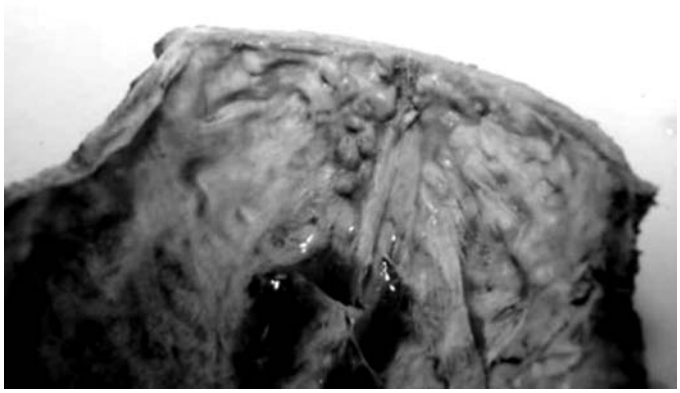

FIG. 1—*Case 2. General view of the inner table of the frontal bone, showing typical location of HFI.*

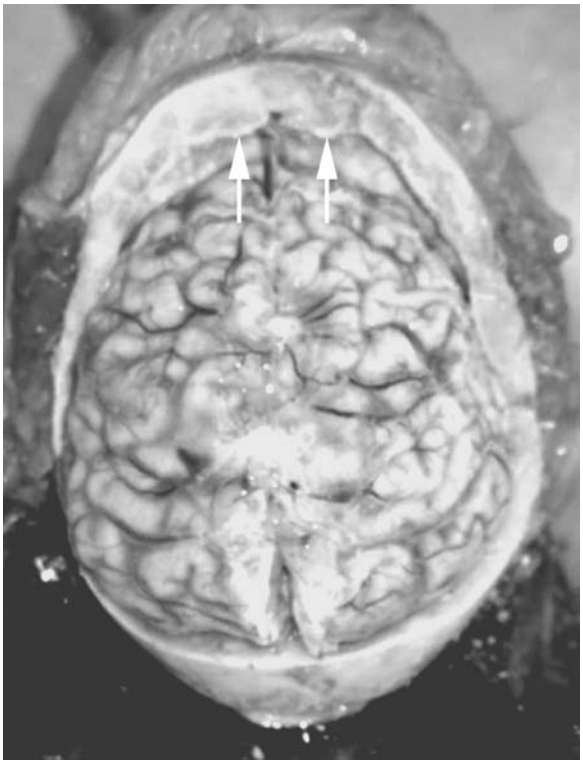

FIG. 2—*Case 7. Type D HFI (classification of Hershkowitz et al., 1999), showing marked thickening of the inner table of the frontal bone.*

with hyperostosis cranialis diffusa that is a generalized thickening of the calvaria with absence of nodules on the inner Table 1.

On routine autopsy radiographs (frontal view), frontal hyperostosis could clearly be suspected in 8 cases (Fig. 3). The accompanying symptoms observed and the causes of death are reported in Table 1.

The accompanying symptoms most frequently found in our sample were behavioral disturbances, aggression or psychiatric disorders, present in 12 individuals of whom over half were under regular psychiatric care. Although the information we had concerning the autopsied persons was not always exhaustive, as far as we know about 30% of our study sample was or had been under medical care for various psychiatric disorders. The remaining individual had no known psychiatric history but suffered from chronic alcoholism. The second symptom was male-pattern obesity that affected 9 of the 13 subjects, and 4 subjects presented Morgagni-Stewart-Morel syndrome.

Discussion

Non-Recognition of the Early Stages of HFI Leads to Underestimation of Incidence

When hyperostosis frontalis interna is particularly well developed, its specific characteristics allow an unequivocal diagnosis to be made both on gross examination and on radiographs (Fig. 3). On the other hand, in the very early stages, it is very difficult to clearly identify HFI because of the existing differential diagnosis. In fact, the bony overgrowths which are small in size and sometimes isolated—types A and B according to Hershkovitz et al. (1)—are very discrete and are often considered as anatomic variations or non-specific endostosis (1). Similarly, it is difficult to identify these types of HFI on radiographs because on an anterior projection the cranial structures are superimposed, and so we could not always make the diagnosis on the basis of radiographs alone. HFI is characterized by thickening of the frontal bone associated with roughness of the endocranial surface. It then resembles a homogeneous mass of dense bone, extending from the outer surface of the skull to

TABLE 1—*Associated symptoms and causes of death in individuals with HFI (PD: psychiatric disorder as depressive and/or frontal syndrome).*

Case	Age	Sex	Morphology	Brain (weight, g)	Features	Context	Cause of Death	Association	Histology
1	49	F	M	1423	–	obesity	brain hemorrhage	arterial hypertension	+arteriolosclerosis
2	42	F	F	1000	asymmetric cerebral atrophy	PD (treated)	pulmonary embolism	–	atrophy
3	71	F	F	lysis	–	PD (untreated)	diabetes	–	–
4	59	F	N	1166	pituitary hypertrophy	obesity + PD (untreated)	epilepsy	warts and virilism	–
5	52	F	F	lysis	–	PD (treated)	ruptured aneurysm	virilism	–
6	74	F	M	1100	pituitary hypertrophy	PD (untreated)	ruptured aorta	atherosclerosis	–
7	66	F	F	1022	–	PD (treated)	strangulation	–	calcifications
8	79	F	M	1214	–	senile dementia	diabetes	–	–
9	51	F	M	1230	–	obesity + PD	diabetes	atherosclerosis	–
10	53	F	M	1150	–	PD (treated)	fall	virilism	–
11	43	M	M	1390	–	PD (treated)	drowning	height 205 cm	–
12	72	F	M	1020	–	senile dementia	road traffic accident	–	Alzheimer
13	58	F	M	1110	–	PD (untreated)	fall	virilism	–

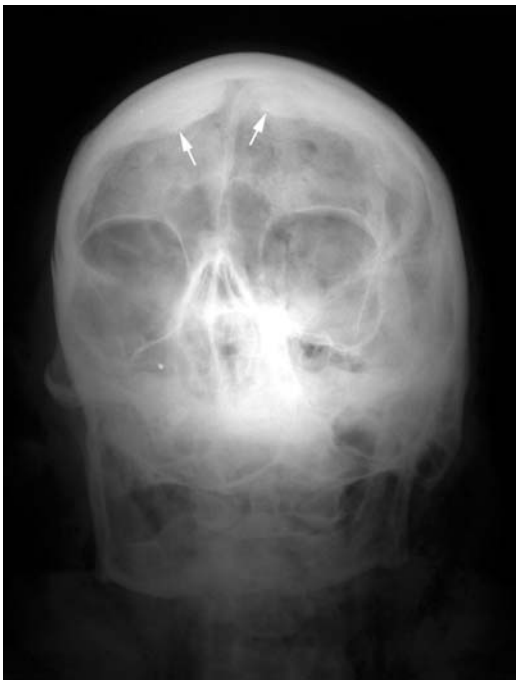

FIG. 3—*Case 5. Anterior radiograph showing the development of HFI on each side of the crista frontalis.*

the limits of the hyperostotic area. Identification of minimal bony overgrowths requires an oblique radiograph, which is not part of standard autopsy procedure.

This latter point no doubt leads to considerable differences in the estimations that have been made up to now, inasmuch as numerous authors have based their studies on radiological examination. It is probable that our own study is not exempt from this problem. The total absence of types A or B HFI in our clinical sample does not seem to reflect reality. According to the study of Hershkovitz et al. (1), about 64% of HFI were types A or B in a total of 219 cases (all ages and sex taken together).

HFI is still a relatively little known phenomenon, notably when it presents as small isolated bony overgrowths, and this certainly has an impact on evaluation of its incidence in autopsy populations.

Incidence of HFI in Autopsy Populations

Only 13 cases of HFI were identified among the 1532 autopsies carried out in the medico-legal department, yielding an incidence of 0.8%. Compared with the most recent publications, this figure seems low. Indeed, according to Hershkovitz et al. (1), the incidence of HFI in the general population may be 12.8%. This figure in fact varies greatly according to the publications. While Moore (2) found an incidence of 1.2%, Grollman and Rousseau (3) advanced a figure of 4.1%, whereas Marlet (4) estimated the incidence at 11.5%. It is probable that this variation arises from the problem we have already mentioned concerning the identification of the early stages of HFI (types A and B), of which only rare examples have been described in the literature.

Influence of Sex and Age

Our findings agree with all published data concerning the gender-related predominance of HFI, confirming that the vast majority of individuals affected are women. We agree with Ortner (5) and Brogdon (6) that this is a very interesting parameter that should be taken into consideration in medico-legal identification.

Looking at the literature, age seems to be strongly linked with the incidence of HFI. Among the 13 cases that we identified in our study population, no individual was aged less than 40 years old. Although some cases of young subjects with HFI have been published (Grollman and Rousseau (3) reported that the youngest patient in their sample was only 21 years old, Cocheton et al. (7) observed a man aged 21 with HFI, and Marlet (4) found two cases of women aged between 15 and 19 years in a total of 300 women), HFI appears to affect principally women of mature age.

Associated Symptoms and Their Implication in Cadaver Identification

The etiology of HFI remains unknown. Several hypotheses have been put forward: HFI could result from prolonged estrogen stimulus during the reproductive period (1) or from disturbances of gluco-regulation (8), but according to some authors it is more likely to be linked with obesity (7), arterial hypertension (2,9) or to have a genetic origin (10,11). In our study, three cases presented vascular lesions resembling arteriolosclerosis or atherosclerosis, but such lesions are frequent in individuals aged over 50 years.

There may be associated menstrual disorders, virilism, hirsutism, mental disorders, fatigue, somnolence, irritability, aggressive behavior, anxiety, visual disorders, vertigo, tinnitus, obesity, polyphagia, polydipsia, polyuria, loss of sense of smell, decrease in glucose tolerance, convulsions, and involvement of the second, fifth, and seventh cranial nerves with hemiplegia and hemiparesis. Diseases that may be associated are Pick's disease, Alzheimer's disease, cerebral atherosclerosis and dementia praecox (9–10).

Although numerous associations of symptoms have been observed since the 18th century, no causal relationship has been revealed. Most studies are therefore based on statistical analysis of large samples, since it has not been possible to clarify the links between the bony lesions as such and the symptoms that frequently accompany them. The symptoms most frequently mentioned in the literature as being associated with HFI are obesity, generally of masculine morphological type (also known as android obesity), virilism (male-pattern hirsutism, well-built face and frontal sinus development) and psychiatric disorders, classified together under the nosological term of Morgagni-Stewart-Morel syndrome. Among these three associations, behavioral disturbances and obesity were the criteria most frequently met with in our sample, followed by virilism. Behavioral disturbances appear strikingly frequent in observations of HFI. Some authors demonstrated that the incidence of HFI was higher in psychiatric hospitals than in general hospitals (12) and the pathologic pattern described was frontal syndrome, which implies abnormal emotional behavior, disturbed reasoning and judgment, and uninhibited reactions. All the individuals in our sample did in fact seem to be affected by some of these problems. So while these various symptoms are not found with the same frequency in all publications or samples, they are cited sufficiently often to be accorded medico-legal value.

Conclusion

In this study, we determined the incidence of hyperostosis frontalis interna as revealed by medico-legal autopsies. We also described the associated symptoms and discussed the fact that the observation of HFI is an important element in medico-legal identification; on the one hand, it contributes to individual identification

by allowing comparison with the elements of the judicial inquiry and on the other hand, it reinforces the reliability of sex and age determination by classic methods. It is therefore necessary to be familiar with this condition and with the different forms it may take. While the symptoms associated with this condition are not found with the same frequency in all publications, obesity, virilism and behavioral disturbances are regularly cited. Hyperostosis frontalis interna should therefore be considered as a supplementary factor in the elaboration of hypotheses relating to an unidentified individual, in particular in a skeletal state.

References

1. Hershkovitz I, Greenwald C, Rothschild BM, Latimer B, Dutour O, Jellema LM et al. Hyperostosis frontalis interna: an anthropological perspective. Am J Phys Anthropol 1999;109:303–25. [PubMed]
2. Moore S. Calvaria hyperostosis and the accompanying symptom complex. Arch Neurol Chicago 1936:35:975.
3. Grollman A, Rousseau JP. Metabolic craniopathy: a clinical and roentgenologic study of so-called hyperostosis frontalis interna. JAMA 1944;126:213–7.
4. Marlet JJ. Development of cranial hyperostosis. Radiol Clin Biol 1974;43:473–82. [PubMed]
5. Ortner DJ, Putschar WG. Identification of pathological conditions in human skeletal remains. Washington, D.C.: Smithsonian Institute Press, 1985;294.
6. Brogdon BG. Radiological identification: anthropological parameters. In: Brogdon BG editor. Forensic radiology. Boca Raton, FL: CRC Press, 1998;75.
7. Cocheton JJ, Fineltain L, Poulet J. Le syndrome de Morgagni-Morel, mythe ou réalité? Sem Hop 1974;50(47–48):2945–50. [PubMed]
8. Mayer R, Martin E. Le syndrome de Morgagni-Morel (un état de diabète latent). Journées annuelles de diabétologie de l'Hôtel-Dieu, Paris 1969:77–94.
9. Mayer R. Hyperostose frontale interne. Analyse statistique, anatomopathologique et clinique d'une série de 100 cas. Acta Neuroveg (Wien) 1962;23:393–413. [PubMed]
10. Rosatti P. Une famille atteinte d'hyperostose frontale interne (syndrome de Morgagni-Morel) à travers quatre générations successives. J Genet Hum 1972;20(3):207–52. [PubMed]
11. Klein D, Rosatti P, Mach RS, Ferrier PE, Rauch S. Le syndrome de Morgagni-Morel, une entité clinique et génétique indépendante, démontrée par une observation familiale s'étendant sur quatre générations. Arch Suisses Neurol Neurochir Psychiatr 1973;112(2):239–50.
12. Hawkins TD, Martin L. Incidence of hyperostosis frontalis interna in patients at a general hospital and at a mental hospital. J Neurol Neurosurg Psychiatry 1965;28:171–4. [PubMed]

Additional information and reprint requests:
Piercecchi-Marti, M.D.
Laboratoire de Médecine Légale
Faculté de Médecine de Marseille
27 Boulevard Jean Moulin
13385 Marseille Cedex 05
France