

Open Access

Hyperostosis frontalis interna: A case report

Adegbenro Omotuyi John Fakoya^{1*}, Jessica Heymans², Amanda McCrary², Omar Rodriguez², Abdiel Cardona², Abayomi Gbolahan Afolabi¹, Thomas McCracken¹

¹Department of Anatomy, University of Medicine and Health Sciences, Basseterre, St. Kitts and Nevis, ²University of Medicine and Health Sciences, Basseterre, St. Kitts and Nevis

ABSTRACT

Hyperostosis frontalis interna (HFI) is a rare disorder common among postmenopausal females that involve increased volume and porosity of the frontal bone. Depending on the size, it could be symptomatic or asymptomatic. With a little report on the pathophysiology of HFI, we present a peculiar case of HFI with speculations that could shed more light on the already established literature and pose questions for future research.

Keywords: Hyperostosis frontalis interna; postmenopausal; frontal bone; diploe plate

INTRODUCTION

Hyperostosis frontalis interna (HFI) is an abnormal growth of the frontal bone at the anterior part of the skull (1). It is characterized by an increased volume and porosity of the inner and diploe plate of the frontal bone (2). Although there is increased volume, due to the porosity, there is no increase in the actual amount of bone tissue (1). The finding is not visible externally but is detectable by an X-ray scan or at postmortem examination, as in our case. The gross changes were evident for the medical students to find, and it seemed unlikely that such a space-filling lesion would be asymptomatic. The literature review and case report presented here aims to identify cases where symptoms have been identified and pose questions for further study.

DOI: https://doi.org/10.17532/jhsci.2020.903



HFI is most predominant among females, with a higher incidence among postmenopausal women (2). It is suggested that HFI is associated with hormonal changes that lead to angiogenesis, causing changes in the frontal bone structure (3). Clinical symptoms attributed to HFI are nonspecific and remain limited to case reports and include neuropsychiatric symptoms (4).

Previous studies have suggested various probable causes of HFI and associated symptoms (5-7), but these are rather speculative, and further studies and experimental evidence are needed for a solid conclusion. In this study, we present a female cadaver with prominent visible HFI and discuss possible clinical symptoms based on its presentation.

CASE PRESENTATION

The cadaver presented here is from a postmenopausal, normal weight Caucasian woman. The external skull and overlying tissue showed no gross abnormalities. Upon removal of the top portion of the calvarium, a thickened frontal bone and anterior

© 2020 Adegbenro Omotuyi John Fakoya, et al.; licensee University of Sarajevo - Faculty of Health Studies. This is an Open Access article distributed under the terms of the Creative Commons Attribution License (https://creativecommons.org/licenses/by/4.0/), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

^{*}Corresponding author: Dr. Adegbenro Omotuyi John Fakoya, Department of Anatomical Sciences, University of Medicine and Health Sciences, Basseterre, St. Kitts and Nevis. E-mail: afakoya@umhs-sk.net

Submitted: 24 March 2020/Accepted: 24 June 2020



FIGURE 1. Images of the cadaver from left to right: Superior view of the external calvarium, notably without external abnormalities. Inferior view of the calvarium with a clear lambdoid suture posteriorly, and thickened, bubbled frontal bone, and porous cavities are seen in the cutaway. Right-sided view showing the thickness of the frontal bone compared to the much thinner adjacent parietal bone.

parietal bone was discovered. The bone had numerous porous cavities ranging in size from a few millimeters to 3 cm in diameter. The internal surface of the frontal bone also had a bubbled appearance, similar to the shape of the adjacent brain tissue, as shown in Figure 1. It was also observed that the hyperostosis finding ended at the superior sagittal sinus and was limited to the frontal bone only. The sinuses were not involved.

DISCUSSION

HFI is a condition described as a thickened frontal bone, which is divided into two areas: Squamous and orbital. Although HFI predominates among postmenopausal females, just as it was in our study, it is not exclusive to the elderly as it has also been identified in young children after severe head trauma (1,4), as well as frequent presentation in patients with Morgagni, Stewart-Morel, and Troell-Junet syndromes. Bracanovic et al. proposed a new system to classify HFI into two categories based on the differences in the diploe and the inner table width into severe (formerly Class D) and moderate (formerly Classes A, B, and C) (1). The article is most important for proposing a new classification of HFI based on micro-CT images, which showed the previous method of categorizing HFI based on the percentage of the frontal bone affected and the number/size of pores to be invalid from a microscopic standpoint. Their evidence, which is based on the

differences between the control group and the HFI group, reinforces the hypothesis from earlier research that suggests the bone formation seen in HFI arises from increased angiogenesis. Angiogenesis is spurred by estrogen-based growth factors in dural arteries, running through the diploe and inner table (1).

We speculate that based on our observation, the hyperostosis caused pressure on the frontal lobe of the brain. In a study previously done, it was shown that this could cause significant difficulty in carrying out tasks that require active manipulation of information in spatial and nonspatial memory (5). A case of HFI reported symptoms of atypical and non-progressive parkinsonism and depression, both of which were unresponsive to medications (8). As summarized in the report, identifying scenarios of symptomatic HFI is essential in diagnosing and treating, to avoid unnecessary medical interventions (8). It is not possible to rule out these diseases in our specimens. However, the cause of death in the cadaver was unrelated to any neuropsychiatric causes.

Another study described a case of HFI in a woman with schizoaffective disorder. The report suggests that even when the condition is not directly symptomatic, it is associated with many disorders, most notably diabetes (6). In addition, the presence of hyperostosis frontalis may lead to decreased intracranial volume, suggesting a role in dementia (7). Identifying HFI in patients, therefore, may play an important role in the overall health picture of patients.

One curious point to note is that HFI is similar to hyperostosis parietalis interna, but both develop from different embryological tissue. The frontal bone develops from neural crest cells, while the parietal bone develops from paraxial mesoderm (2). As to why one of them is observed in an individual and not the other may lead us to speculate that there are embryological and genetic differences in the two groups of patients with these disorders. Further studies on embryological development, growth factors, angiogenesis, prevalence, and comorbidities are needed to elucidate the causes and associations more clearly.

CONCLUSION

HFI is an uncommonly diagnosed condition associated with increased volume and porosity of the inner and diploe plate of the frontal bone. The case presented here clearly shows the gross changes apparent during the postmortem examination. The literature reports frontal bone hyperostosis occasionally being symptomatic and associated with comorbidities, so more research might prove helpful in treating therapy-resistant neuropsychiatric disorders.

REFERENCES

 Bracanovic D, Djonic D, Nikolic S, Milovanovic P, Rakocevic Z, Zivkovic V, et al. 3D-microarchitectural patterns of hyperostosis frontalis interna: A micro-computed tomography study in aged women. J Anat 2016;229(5):673-80.

https://doi.org/10.1111/joa.12506

 Western A, Bekvalac J. Hyperostosis frontalis interna in female historic skeletal populations: Age, sex hormones and the impact of industrialization. Am J Phys Anthropol 2016;162(3):501-15.

https://doi.org/10.1002/ajpa.23133

 Brandi M, Collin-Osdoby P. Vascular biology and the skeleton. J Bone Miner Res 2006;21(2):183-92.

https://doi.org/10.1359/jbmr.050917

- Li Y, Wang X, Li Y. Hyperostosis frontalis interna in a child with severe traumatic brain injury. Child Neurol Open 2017;4:2329048X1770055. https://doi.org/10.1177/2329048x17700556
- de Zubicaray G, Chalk J, Rose S, Semple J, Smith G. Deficits on self ordered tasks associated with hyperostosis frontalis interna. J Neurol Neurosurg Psychiatry 1997;63(3):309-14. https://doi.org/10.1136/jnnp.63.3.309
- Chaljub G, Johnson III R, Johnson R Jr., Sitton C. Unusually exuberant hyperostosis frontalis interna: MRI. Neuroradiology 1999;41(1):44-5. https://doi.org/10.1007/s002340050703
- May H, Mali Y, Dar G, Abbas J, Hershkovitz I, Peled N. Intracranial volume, cranial thickness, and hyperostosis frontalis interna in the elderly. Am J Hum Biol 2012;24(6):812-9.

https://doi.org/10.1002/ajhb.22325

 Batun GC, Yuruyen M, Vatankulu B, Elmali A, Emul M. Hyperostosis frontalis interna presenting as depression and parkinsonism in an older woman. Psychogeriatrics 2015;16(5):329-30.

https://doi.org/10.1111/psyg.12166