Hyperostosis frontalis interna associated with hypogonadism in an elderly man

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Abstract

Hyperostosis frontalis interna (HFI), symmetric thickening of the inner table of the frontal bone, is relatively common in women but very rare in men. We report the case of an elderly male patient with HFI. This patient was accompanied by primary hypogonadism, which may be related to the underlying pathogenesis of HFI.

Keywords: hyperostosis frontalis interna, frontal bone, male, primary hypogonadism, elderly

Introduction

Hyperostosis frontalis interna (HFI) is a condition involving limited proliferation of the inner table of the frontal bone, which may be accompanied by endocrinopathies (e.g. myxoedema [1] and hyperprolactinaemia [2]) and/or neuropsychiatric abnormalities (e.g. schizoaffective disorder [3] and behavioural disturbance [4]) in some cases. Although the aetiology of HFI remains unclear, there is marked female predominance [4–6] in the epidemiology of this disease. Here, we present the case of a rare male patient with HFI. Interestingly, this case was accompanied by hypogonadism.

Case report

A 72-year-old man (height 178 cm and weight 73 kg) developed right occipital infarction 1 month before admission to our hospital for rehabilitation. On examination, left homonymous hemianopia and left hemiparesis were noted. Cranial computerised tomography (Figure 1) and magnetic resonance imaging showed HFI in addition to an ischaemic lesion in the right occipital lobe. Subsequently, we noted that his penis was small without palpable testes. He had never needed to shave and axillary hair was absent. Gynaecomastia was not noted. The patient and his wife had no children, although he was not impotent at least in his early years. Laboratory testing demonstrated a markedly reduced level of testosterone (21.3 ng/dl; normal range for males, 270–1,070 ng/dl) and elevated levels of luteinising hormone (42.6 IU/l; normal range for males, 1.2–7.1 IU/l) and follicle-stimulating hormone (26.9 IU/l; normal range for males, 2.0–8.3 IU/l), which were consistent with primary hypogonadism (hypergonadotropic hypogonadism). We could not perform further investigations including chromosome analysis because the patient suddenly died of acute respiratory failure following a further cerebral infarction.

Discussion

Despite marked female predominance in HFI, masculinisation has been observed in some female cases [4, 5]. In addition, Kollin and Fehér [7] have shown that HFI is related to raised androgen levels in premenopausal women. These findings suggest that gonadal imbalance may underlie the disease pathogenesis. On the other hand, Hershkovitz et al. [8] reported a male case of HFI with a single atrophied testis in cadaver studies. In that article, they pointed out that male cases of HFI with feminisation and/or testicular atrophy had been described in earlier reports from the mid-twentieth century. In addition, there are reports of HFI associated with Klinefelter’s syndrome [9] and Kallmann syndrome [10]. Taken together with these previous observations, our case indicates that underlying conditions should be checked if HFI is detected in a male.
Key points

- Endocrinopathies and/or neuropsychiatric abnormalities may accompany HFI in some cases.
- There is marked female predominance in the epidemiology of HFI.
- Hypogonadism should be checked if HFI is detected in a male.

References


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