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Rare association of diffuse idiopathic skeletal hyperostosis and hyperostosis frontalis interna

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Abstract
Published case studies of the association of diffuse idiopathic skeletal hyperostosis (DISH) and hyperostosis frontalis interna (HFI) are rare. In this manuscript, we report a case of dorsal spine HSID in a 57-year-old man and HFI with spinal pain and headache to discuss the etiopathology, the key imaging and clinical features of these conditions after literature review.

Keywords: diffuse idiopathic skeletal hyperostosis, hyperostosis frontalis interna, CT scan

1. Introduction
DISH is a systemic disorder characterized by ossification of vertebral ligament and peripheral enthesis [1]. The hyperostosis frontalis interna (HFI) was described for the first time by Morgagni in 1751 characterized by a thickening of the internal table covered by the dura mater, protruding into the cranial cavity [2].

The prevalence of DISH is higher in men (generally in their 60s) than in women. Incidence increases with age and varies by population. In contrast, HFI have been reported in 5 to 12% of the general population, they are much more common in older women [3].

2. Observation
A 57-year-old man was referred to the radiology department of Antsiranana University Hospital for mechanical backbone pain and persistent headache. On anamnesis, he is diabetic type 2; his body mass index is 32. The serum uric acid level was 90 mg / dl. The CT scan showed a continuous bone flow in the dorsal segment that borders the anterior surface of the thoracic vertebral bodies from T4 (Figure 1, 2), the intervertebral discs were not pinched, and there was no evidence of abnormalities of the vertebral trays; smooth-surface thickening of the frontal, bilateral and symetrical thickening of the inner table of the frontal bone (type D) with calcification of the skull of the brain (figure 3).

3. Discussion
DISH is a non-inflammatory systemic disease of unknown etiology. All authors agree on its multifactorial character.

The mechanical factors leading to ligament stretching have been questioned. Diabetes, obesity, dyslipidemia and hyper-uricemia have been reported in patients with DISH [4]. Exposure to certain substances such as fluoride, vitamin A / retinol; drugs in this case isotretinoin, etretinate, acitretin and other vitamin A derivatives have been described [5].

Currently, most theories focus on the pathological calcification of anterior vertebral ligament. Most of these theories postulate that this process is due to abnormal growth secondary to osteoblastic hyperactivity, so there is an association between this pathological calcification and the increase in bone mineral density [6].

When with internal frontal hyperostosis, the exact etiology is still unknown. There are endocrine factors and environmental factors. The prevalence of the disease increases with age and is more common in women; it goes from less than 10% in women under 30, to almost 60% in women over 60, so after menopause [3]. Other factors have been shown such as diabetes mellitus, advanced age, morbid obesity and diet, coronary artery disease and hypertension as factors of development of HFI. A prevalence of 84% HFI was observed in obese subjects (> 40% of ideal body weight), while in thin subjects, HFI was present in only 16% of cases [7].

DISH and HFI, both found in this analyzed individual, are characterized by abnormal bone growth, late onset of age, and association with obesity and various metabolic disorders. In their study, Wilczak and Mulhern [8] examined the co-occurrence of DISH and HFI in one study. DISH was present in 20% of black men and 27.6% of white men with HFI, but it was not significantly different from that of male controls without HFI (16.8% and 18.9%). They suggest that similar metabolic, genetic, and environmental risk factors in the pathogenesis of both conditions explain the observed comorbidity.

DISH affects men over 50 years old. Its incidence increases with age. 13 to 32% of patients have diabetes associated with this disorder. It is an often asymptomatic disease [9]. Frequent symptoms are pain like that of our case, stiffness and reduced mobility of affected areas. Complications should be suspected in case of acute symptoms. The most severe are cervical myelopathies, vertebral fractures and compressions of other structures (esophagus, larynx and trachea, inferior vena cava...) [10]

HFI has particularly affected women in the postmenopausal period [3], which is not our case.

The clinical manifestation of HFI is variable. HFI is usually asymptomatic but may also be associated with headache, which is found in our case, cerebral atrophy or neuropsychiatric manifestations, epilepsy, or dementia [11].

As these diseases are most often asymptomatic, they are fortuitously discovered on imaging tests. It is a disease that ossifies the entheses, ligaments and joint capsule. The most commonly used classification criteria were defined by Resnick and Niwayama and required after anterolateral ossification: at least four contiguous thoracic vertebral segments, preservation of intervertebral disc spaces, and absence of apophyseal joint degeneration of the joints or sacral inflammatory changes iliacs [12].

After the study conducted by Kim *et al*, the most common location of DISH was the average thoracic spine (90.0%), followed by the lower thoracic spine (87.5%) [13].

The HFI is manifested as an irregular, partly nodular, thickening of the internal table of the voice of the skull. The bone structure is otherwise normal, it is neither destroyed nor modified. The HFI is classified according to its severity (shape and size) into four types [14]:

- Type A: isolated bone elevations of the endocranial surface of diameter less than 10 mm.
- Type B (minor HFI): bone growths occupying less than 25% of the frontal bone area.
- Type C (moderate HFI): larger bony outgrowths occupying up to 50% of the frontal bone area.
- Type D (severe HFI): bone growths involving more than 50% of the frontal bone surface

4. Figures

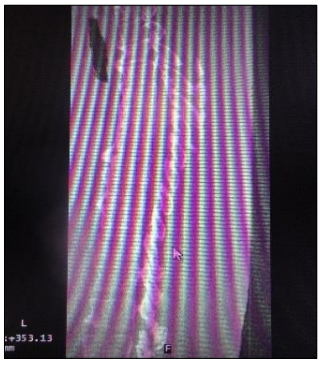

Fig 1: Sagittal dorsal CT scan – Bone window

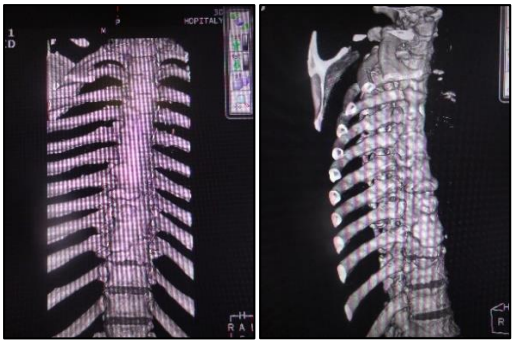

Fig 2: CT scan in 3D reconstruction, coronal and lateral view, showing waxy flowing appearance of the candle

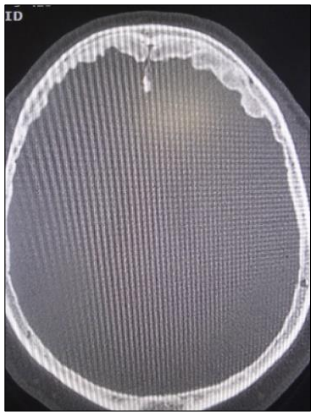

Fig 3: Bilateral thickening of the inner table of the frontal bone

5. Conclusions

DISH and HFI are the preserve of the elderly. The association of these two affections is rare. Several hypotheses have been put forward, but their causes are still unknown. It is possible that similar metabolic and environmental risk factors in the pathogenesis of both conditions may explain the observed comorbidity. The diagnosis is always radiological.

6. References

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