Hypertrophic Osteoarthropathy: A Rare Manifestation of an Amygdalar Tumour

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INTRODUCTION

Hypertrophic osteoarthropathy (HOA) is a syndrome that associates the characteristic deformity of the finger tips and periosteal proliferation of the long bones. The syndrome has been known since the last century by the name of von Bamberger Pierre-Marie1. It may present as primary HOA, as in the case we present, or secondary HOA, generally associated with pulmonary, cardiovascular, and infectious (including the human immunodeficiency virus [HIV]) processes, carcinomas of the nasopharynx and epidermoid carcinomas of the lung with hormonal production and even neoplasms of the larynx2. It has also been related to hormones that foster periosteal stimulation and other factors continue to be under study that might favour its origin.

When faced with a case that is compatible with HOA, we are obliged to rule out underlying oncological diseases that tend to appear after the initial lesion and are generally related to pulmonary metastases. It is currently considered a true paraneoplastic syndrome3. In the area of otorhinolaryngology, it has been related to tumours of the cavum, with pulmonary metastatic potential, particularly in young patients3–5.

We report on the case of a patient in whom the condition was detected months before the appearance of a malignant amygdalar tumour and in whom the osteoarticular process improved following treatment of the neoplasm.

CLINICAL CASE

A 46-year old patient, with a personal history of significant smoking and alcoholism, no prior rheumatic or allergic history who presented at the orthopaedic outpatient clinic due to pain in the right ankle without any clear history of trauma, non-radiated cervical pain. Following a general radiological work-up, the patient was diagnosed as a carrier of degenerative phenomena. In light of the lack of response to anti-inflammatory treatment,
the decision was made to carry out a bone gammagraphy, which revealed an area of intense, abnormal bone tracer uptake in the distal portion of the left peroneal diaphysis, above the malleolus and compatible with a focus of pseudoarthrosis, intense, irregular increased uptake in both femurs and the right tibia with clear cortical reinforcement, a gammagraphic pattern that was highly suggestive of a pneumic HOA metabolic process. The patient was sent to the department of rheumatology for study of these alterations (Figure 1).

Physical examination: good general status, gets good exercise, normal colour, no fever, cardiac and respiratory auscultation unremarkable, no visceromegalies. Prior to completion of the work-up, the patient reports amygdalar discomfort and is referred to the otorhinolaryngology service, where a right amygdalar neoformation and adenopathy in the right jugulogastric space are detected (T2N1). Positive amygdalar biopsy for keratinizing squamous cell carcinoma (Figure 2). Laboratory tests: haemogram, transaminases, kidney function, haemostasis, glycaemia, alkaline phosphatases and other biochemical parameters, normal. Hepatitis B virus, hepatitis C virus, and HIV serology, negative. Normal venous gasometry. Chest film, normal.

After examination by the anaesthesia service, the patient underwent surgery including a broad tonsillectomy and bilateral functional node excision. Subsequently, the patient underwent complementary physical treatment, according to the centre’s protocol.

Pathology examination: well-differentiated, keratinizing epidermoid type carcinoma, infiltrating the amygdalar parenchyma. In the right node basin, a node is seen to be infiltrated with squamous cell carcinoma (1/12) (Figure 3).

**DISCUSSION**

Many malignant neoplasms have been related to paraneoplastic rheumatic syndromes such as dermatomyositis/polymyositis, paraneoplastic vasculitis, carcinomatous polyarthritis, and HOA. HOA is defined by the presence of deformities of the distal areas of the extremities in the form of clubbed or “Hippocratic” fingers, with proliferation of the periostium of the long bones (Figure 4). Most patients are asymptomatic or they may complain of pain in the extremities. An important trait detected during examination is the presence of clubbed fingers. The ankle may be an area presenting periostitis, painful upon palpation that is readily distinguishable from arthritis pain. X-ray is a useful means to detect abnormalities and periosteal...
Juxtaposition can be seen in the diaphysis that is symmetric in the long bones. Bone gammagraphy is a sensitive method to demonstrate the periosteal condition and can reveal lesions before X-rays. It is not specific to cancers, given that it can be seen in the course of cyanogenic cardiopathies, chronic inflammatory liver disease; however in 90% of the cases, it is secondary to a tumour inside the chest: primary or secondary lung tumour (in 75% of cases), pleural mesothelioma, mediastinal tumour or most exceptionally, in malignant blood disorder or in otorhinolaryngological cancer. Generally tumours of the nasopharynx in young people and even in the maxillary sinus and in the larynx, although in the case reported in the medical bibliography, the patient also had simultaneous pulmonary carcinoma.

HOA and the neoplastic process present in variable ways, usually the tumour already exists when HOA manifests, albeit in some cases, HOA can precede the diagnosis of the tumour or as an early indicator of metastases, usually pulmonary metastases. In the case we report, the osteoarticular symptoms appeared several months prior to the diagnosis of the tumour. Of the tumours of the oral cavity, amygdalar tumours represent 50% and tend to be epidermoid; they have been associated with poor oral hygiene, diabetes, granulomatous diseases and alcohol/tobacco use. Approximately 56% of patients present with positive nodes. Diagnosis will be histopathological and the first treatment option is surgery of the tumour and cervical nodes. The evolution of osteoarthropathy tends to respond favourably to the treatment aimed at the underlying cancer and often disappears. The main pathogenic factor of HOA would be increased peripheral circulation of the affected joints, although many other factors have also been posited.

The clinical case presented is the first case so far, according to the bibliography consulted, to relate both conditions. As a conclusion, we could say that the association of HOA with malignant neoformations is common and the tumour causing the condition must be intensively sought; it need not be located inside the chest or cavum (rhinopharynx), since other otorhinolaryngological locations cannot be ruled out, as commented in the article. Where the tumour is known, HOA may help us predict the metastasic spread of the initial tumour.

REFERENCES