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ORIGINAL ARTICLE

Sterno-clavicular prominence: hyperostosis or interarticular ossifiction. It is a ragbag?

C. Villas-Tomé,* P. Díaz-de-Rada, L.M. Romero-Muñoz, A. Valentí-Azcárate

Departamento de Cirugía Ortopédica y Traumatología, Clínica Universidad de Navarra, Facultad de Medicina, Universidad de Navarra, Pamplona, Navarra, Spain

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KEYWORDS
Sterno-clavicular hyperostosis;
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Abstract
Introduction: In clinical practice, we see quite often painful sterno-clavicular prominence, deformity, which is the hallmark of two entities with a low incidence, although well defined hyperostosis cost sterno-clavicular (HECC) and ossification cost inter-sterno-clavicular (OIECC). The differential diagnosis of these conditions, you must include sterno-clavicular arthritis, Paget's disease, condensing osteitis, osteomyelitis, pustulosis, Friederich's disease, Tietze's syndrome and osteoid osteoma. This is especially important in cases where the involvement is unilateral.
Material and methods: We present a series of nine patients complaining of a painful sterno- clavicular prominence, compatible with HECC or OIECC. The reason for consultation in most patients was rule out the presence of a tumor in that location.
Results: Image studies showed a variable increase both the density and bone mass as well as different intensities of joint involvement sterno-clavicular cost.
Conclusion: Ruled out a tumor and a specific diagnosis of these, you do not need aggressive treatment, is generally considered sufficient anti-inflammatory treatment in most patients. In clinical practice, it is irrelevant and OIECC HECC differentiation, since treatment and prognosis of both conditions are similar. The biopsy of the joint and aggressive diagnostic procedures may be unnecessary.

PALABRAS CLAVE
Hiperostosis esterno-clavicular;
Osificación inter-esterno-costo-clavicular

Prominencia esternoclavicular: hiperostosis u osificación interarticular ¿Un cajón de sastre?

Resumen
Introducción: En la práctica clínica observamos con cierta frecuencia una prominencia esternoclavicular dolorosa, deformidad que constituye el principal signo de dos entidades con baja incidencia, aunque bien definidas: la hiperostosis esterno-costo-clavicular

* Corresponding author.
E-mail: cvillas@unav.es (C. Villas-Tomé).

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(HECC) y la osificación inter-esterno-costo-clavicular (OIECC). El diagnóstico diferencial de estas afecciones debe incluir la artrosis esterno-clavicular, la enfermedad de Paget, la osteitis condensante, la osteomielitis, la pustulosis, la enfermedad de Friederich, el síndrome de Tietze y el osteoma osteoide. Esto es especialmente importante en los casos en los que la afectación sea unilateral.

Material y método: Presentamos una serie de nueve pacientes cuyo motivo de consulta fue una prominencia esterno-costo-clavicular, compatible con HECC u OIECC. El motivo de consulta en la mayoría de los pacientes fue el descartar la presencia de un tumor en esa localización.

Resultados: Los estudios radiológicos mostraron un aumento variable tanto de la densidad como de la masa ósea, así como diferentes intensidades de afectación de la articulación esterno-costo-clavicular.

Discusión: Descartado un tumor y con un diagnóstico concreto de la causa de la prominencia, generalmente, se considera suficiente con el tratamiento antiinflamatorio en la mayoría de los pacientes. En la práctica clínica parece irrelevante la diferenciación entre HECC y OIECC, ya que el tratamiento y el pronóstico de ambas afecciones son superponibles. La biopsia de la articulación y los procedimientos diagnósticos invasivos pueden ser innecesarios.

Introduction

It is not uncommon to see patients in clinical practice who consult for a palpable prominence in the sterno-clavicular area that is usually painful and swollen. Imaging studies of these patients, particularly conventional x-rays, show different pathological images that usually approach the problem as an unspecific rheumatic disease. That is why the following step is usually a biopsy.

There are two clinical entities, sterno-costo-clavicular hyperostosis (SCCH) and inter-sterno-costo-clavicular ossification (ISCCO), which could account for the majority of disorders dealing with an increase in volume in the sterno-clavicular area. Despite this, the diagnosis of these entities is rare. In differential diagnosis, particularly those with unilateral involvement, we should also include Paget's disease, condensing osteitis, osteomyelitis, Friederich's disease, Tietze's syndrome, osteoid osteoma, sterno-clavicular arthritis and other clinical entities.[1-10]

Sterno-costo-clavicular hyperostosis is a rare rheumatic entity first described by Caffey and Silverman in 1945.[4] It is characterised by swelling in the sternal region, clavicle and upper ribs, as well as affecting the thoracic spine. Clinical signs include painful episodes, especially at night, shoulder abduction limitation and even, although less frequently, an increase in collateral venous circulation of the neck. Radiological images show regional bone sclerosis, as well as inter-sterno-costo-clavicular synostosis with ossification in the adjacent soft tissue.[11,12]

Inter-sterno-costo-clavicular ossification was described in 1974 by Sonozaki[13] y Brower.[3] This entity shows the same clinical symptoms but differs in the radiological findings. This entity usually has bilateral affectation and subdivides into three groups. Group 1 presents ossification limited to the costo-clavicular ligament, while group 2 (the most common) presents an extension of ossification on the upper margin of the first rib and on the bottom margin of the clavicle and the space between the first rib and the clavicle is filled with a bone growth. Group 3 is characterised by a hyperostosis of the entire clavicle.

This aim of this work was to analyse clinical and radiological aspects, as well as some physio-pathological findings, in nine patients with pain and deformity in the area and to give some recommendations with regards to diagnosis and treatment.

We hope this article will provide guidance in relating and diagnosing these two entities. We also present a performance guide for patients with sterno-clavicular prominence (sterno-costo-clavicular or sterno-cleido-costal, etc.) in daily clinical practice.

Material and methods

We retrospectively studied 9 patients who visited a doctor between 1979 and 2000 because of an increase in volume or pain in the sterno-costal area and who were assessed by the same medical team during that period. The inclusion criteria for the study were the presence of a prominence located in the sterno-clavicular area and painful swelling in that area. The analytical study had to be normal and simple x-rays had to show hyperostosis of the medial part of the clavicle, sternum and first rib. More than 9 patients with pain in this area were assessed; however, they were excluded as they did not fulfil the study inclusion criteria that formed part of the differential diagnosis.

We collected the physical examination of all patients during their first visit to carry out this study, emphasising whether or not sterno-clavicular prominence was present (fig. 1). The presence of pain, ranges of movement and signs of local swelling were assessed. Radiological studies consisted of x-rays in an anteroposterior and lateral

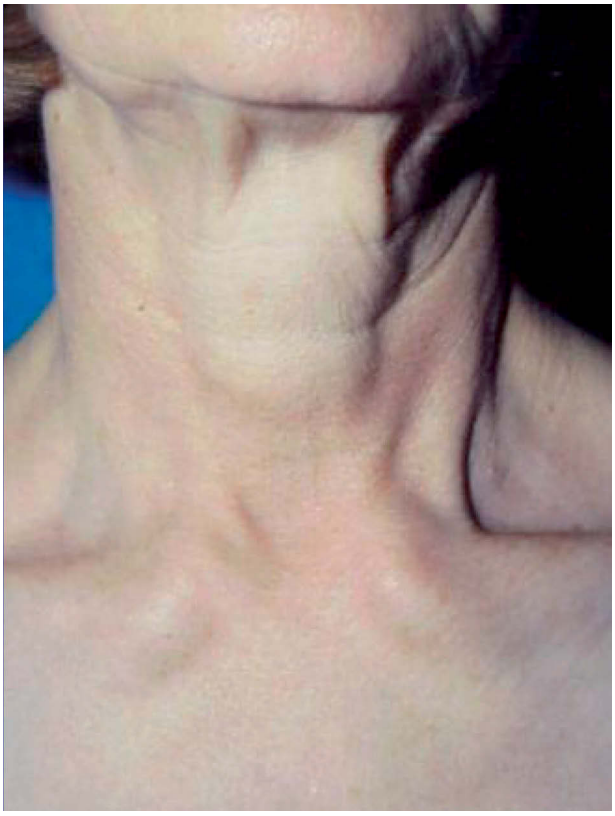

Figure 1 The most frequent finding during examination was uni- or bilateral prominence in the sterno-clavicular area.

projection to see the deformity morphology and delimit the extension of the affectation. Other imaging tests such as computerised tomography (CT) or scintigraphy with technetium were not carried out on all cases, but used only on cases where there was reasonable diagnostic doubt of the first diagnosis. Cases that had an increase in volume were diagnosed with SCCH, and ISCCO was diagnosed in those cases were joint fusion was found.

Results

Seven of the patients were female and 2 were male; the mean age at the time of consultation was 55.8 years (range, 46-63). Four patients presented bilateral affectation. The period from the start of the symptoms to consultation was 22 months.

Eight of the patients consulted because of a painful tumour and swelling in the sterno-clavicular area. One patient consulted for aesthetic reasons; she was completely symptom free at the time of consultation. In the rest of the cases, patients wanted to eliminate the possibility that the increase in volume was secondary to a malignant tumour.

Four patients presented shoulder abduction limitation. Three patients presented signs of local swelling; an increase in collateral venous circulation was not reported in any case.

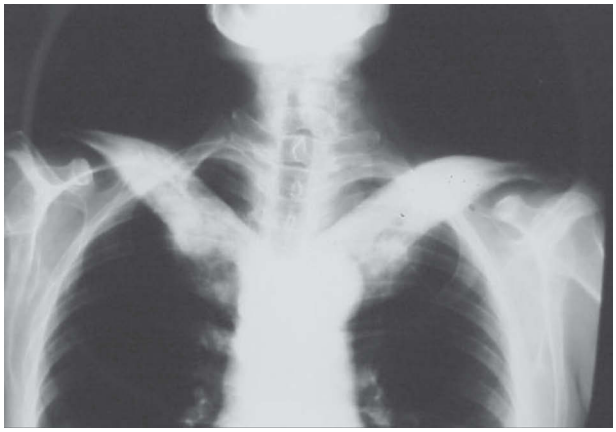

Figure 2 Symmetrical affectation of both clavicles resembles the image of a Spanish goat's horns. The irregular changes of trabecular morphology remind us of that found in Paget's disease.

Laboratory analyses were normal, although increased erythrocyte sedimentation rate (97 and 120 first thing in the morning) and increased Alpha 2 globulins were found in two patients. We could not find an explanation for these analytical changes. No cases of infectious disease were reported in any of the patients.

Imaging studies showed a presence of variable hypertrophy in all cases with greater bone mass, affecting the top ribs (especially the first rib) as well as the bottom of the clavicle, the costosternal area and the sternum. Hypertrophy was so bilaterally marked in one case on the radiological imaging that it reminded us of a "Spanish goat's horns" (fig. 2). This patient also presented affectation in the thoracic spine consistent with decreased disc size and ossification of the longitudinal anterior ligament, in different segments of the upper area. There were four cases of sterno-clavicular hyperostosis, so they were diagnosed with SCCH and the other five were diagnosed with ISCCO.

In cases where a CT scan was carried out, we saw a decrease in joint space, joint surface irregularity, subchondral cysts, osteophytes and bone fusion (fig. 3).

The first three cases of the series had presented an initial diagnosis of Paget's disease, osteoid osteoma and osteomyelitis. In the first case, the scintigraphy showed significant uptake, which made us suspect that it was Paget's disease, but this diagnosis was eliminated by analysis.

In the second case, sclerosis in conventional x-rays, made us suspect it was an osteoid osteoma; this pathology was eliminated by the CT, which did not show images compatible with tumours. The patient presented slight symptom improvement with the use of non-steroidal anti-inflammatory (NSAID) drugs; however, as the discomfort did not go away completely, we carried out a partial clavicle resection, using Kirschner wire as a temporary fixation system. The pathological study of the surgical piece ruled out the possibility of an osteoid osteoma. Based on the pathological findings, we finally diagnosed SCCH. Symptoms improved completely after surgery.

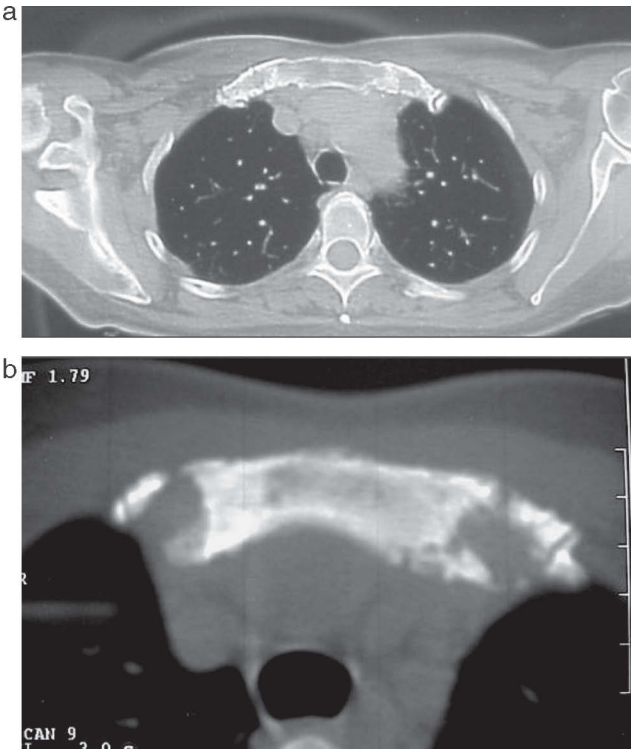

Figure 3 A and B. Hypertrophy and sclerosis in both sterno-clavicular joints.

In the third case, the initial diagnosis at another centre was possible osteomyelitis, because the patient presented pain, signs of local swelling and an increase in temperature, as well as an increase in erythrocyte sedimentation rate. There was complete symptom improvement when NSAIDs were administered.

The use of NSAIDs as a sole treatment had satisfactory results in 6 out of 8 patients who consulted because of pain. Two patients who did not have total improvement of their symptoms with the use of conservative treatment were surgically treated with a partial clavicular resection, with a complete improvement of their symptoms.

In cases where pathological data is available, the pieces were described as trabecular hypertrophy, as well as unspecific inflammatory reaction.

Discussion

Since their description in 1945[4] and 1974,[3,13] there have been few publications on series of patients affected with sterno-costo-clavicular hyperostosis or inter-sterno-costo-clavicular ossification. The few articles published on this subject are also spread out among publications on different specialities. This means that it is difficult to find information, given that specialists in Orthopaedic Surgery rarely look at radiology, internal medicine or plastic surgery magazines. The facts that both entities are not frequently observed and that there is difficulty in finding information in the literature make them unknown affectations for the majority of doctors.[9,10] In practice, being faced with an unknown pathology can lead to unnecessary diagnostic tests (including biopsy).

In our experience, and according to references consulted,[4,5,7,10,12,14-19] SCCH and ISCCO are two entities of unknown aetiology that present predominantly in females and that usually appear in the patients' fifties and sixties. Clinical symptoms include painful swellings located in the sterno-clavicular area, as well as signs of local swelling during acute phases. Some patients presented a slight decrease in shoulder abduction, as well as an increase in collateral venous circulation of the neck. Radiological studies in these patients showed a hyperostosis of the medial part of the clavicle, sternum and first rib. In principle, there are not many diagnostic doubts with this information and, if there are, a CT scan is usually conclusive.

The main difficulty we find when coming across a patient with a sterno-clavicular prominence is to differentiate SCCH from ISCCO of other affectations with similar clinical-radiological manifestations. This is why patients are often submitted to a multitude of diagnostic tests (scintigraphy, CT, etc.), when the definitive diagnosis could be reached with a study of the resected piece. Given that patients affected with SCCH and ISCCO generally respond favourably to taking NSAIDs, it is appropriate to start treatment before undertaking further diagnostic tests or surgery, which would only be indicated if there is a diagnostic doubt or refractoriness to treatment.

In the diagnosis of a patient having sterno-clavicular prominence (painful or not), what is really important is to establish that the patient has no tumours or infection. However, there is a wide range of clinical entities that come under the differential diagnosis. One example is condensing osteitis of the clavicle, an entity that only affects the clavicle and keeps the rest of the bones normally conformed. Half of patients present joint synostosis that includes the first rib.

Paget's disease can produce similar clinical symptoms, but radiological imaging rarely shows joint synostosis. In addition, the patient's analysis would show an increase of alkaline phosphatase not only in the blood but also in urine, as it is an enzyme that maintains normal parameters in patients affected by SCCH or ISCCO. If we biopsy the joint, in Paget's disease there are characteristic intraosteoclastic inclusions that do not appear in these two entities.[3,8,10,14,16] Ultimately, we could also be facing the same entity with a variable clinical and radiographic expression or that could have phases or forms with different radiographic expression. One of our patients, who was initially diagnosed with Paget's disease, presented a bilateral clavicular growth that reminded us of a "Spanish goat's horns". The scintigraphic study showed significant radiotracer uptake in the area where there was an increase in volume.[20] (fig. 4)

A CT scan was carried out on three patients to rule out other affectations such as bone tumours given that, according to some authors,[8] bone metastases and Ewing's sarcoma should be included in the differential diagnosis. We saw obliteration of the intramedular cavity, as well as signs of uniformly sclerosing bone formation in all cases.

Figure 4 Bone scintigraphy showing abnormal radiotracer uptake in both clavicles.

In osteoarthritis, the characteristic data presented are decreases in joint space and the appearance of osteophytes: these are characteristic and differentiating findings from other affectations.[9]

In the case of osteomyelitis, the norm is to see a patient with a history of painful swellings with images where joint destruction and signs of periosteal reaction can be observed. A scintigraphy with gallium or marked leukocytes shows increased uptake. In some cases, the differential diagnosis can be difficult and require a biopsy and culture.[1,2,8,9,14]

Avascular necrosis of the medial epiphysis of the clavicle (Friedrich's disease) is not very common and affects only the clavicle leaving the ribs and the sternum. Radiologically, we see an increase in irregular density of the medial part of the clavicle; definitive diagnosis is only possible with a biopsy.[1,2,8]

Tietze's syndrome is characterised by a swelling of the costosternal cartilages, especially in second rib. Imaging rules it out if there is clavicular affectation, signs of necrosis, sclerosis or joint fusion.[1,2,8]

Patients affected with osteoid osteoma generally present characteristic night-time pain that disappears with aspirin. Imaging tests, especially CTs, should show us the typical nidus image.

To summarise, once the affectations we have detailed in the differential diagnosis have been eliminated, discussion on whether it is SCCH or ISCCO is not very useful because there are two entities with the same symptoms, similar treatment and similar prognosis.[13,16-18,21] Following this principle in clinical practice, treatment and diagnosis of these patients is relatively simple and generally satisfactory (6 out of the 8 patients consulted because of pain), figures similar to other series.[22] In addition, in the majority of these patients we avoided invasive techniques such as biopsy or resection surgery.

Evidence level

Evidence level IV.

Conflict of interest

The authors declare no conflict of interest.

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