CASE REPORT

Xanthoma of the rib without hyperlipoproteinemia

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Summary
Primary bone invasion by xanthoma is rare particularly in normolipidemic patients. In fact, lipid disorders are present in most patients. Xanthomatous lesions were observed in the ninth rib’s posterior arch of a 45-year-old man with a history of labour accident. To our knowledge, this is the fourth case reported in the literature. Histologically, the lesion was characterized by a replacement of normal bone and marrow with xanthoma cells. The treatment approach consisted in a total removal of the lesion and the patient has not presented recurrences after a follow up of 10 years.

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Primary bone invasion by xanthoma is rare. This is especially so in the ribs. These tumours are usually associated to hyperlipoproteinemia. Cases of costal xanthoma in normolipidemic patients are rarely reported in the literature. In the 21 cases of osseous xanthoma reported by Bertoni et al., only three affected the ribs1. To our knowledge, this is the fourth case reported in the literature.

A 45-year-old man with a history of minor trauma from a labour accident presented to his primary care physician because of pain in a left and anterior part of the chest. The patient was in good health and there was no history of obesity, diabetes mellitus or premature atherosclerosis disease. The physical examination was normal. On radiological studies (roentgenogram and CT-scan), the lithic lesion was small (1 cm), unifocal and involved the left ninth rib’s posterior arch without extension to intrathoracic structures or the adjacent soft tissues (Figure 1). The size of the lesion was too small to enable a better analysis. The radiographic appearance was suggestive of a benign process. However, a malignant lesion could not be definitely excluded. The treatment approach consisted in surgical resection. The posterior arch of the ninth rib was totally removed. The operative specimen consisted of a rectangular segment of bone measuring 9 × 2 cm. Macroscopically, a well circumscribed, homogeneous and white yellow tan mass of 1 cm was noted. Microscopically, this mass corresponded to a proliferation of foam cells, cholesterol clefts and fibrosis. This proliferation was well circumscribed and no cytonuclear abnormalities were seen (Figure 2). The surrounding bone was normal, particularly the resected margins. The diagnosis of costal xanthoma was retained. No other additional treatment was indicated. The patient has not presented complications or recurrences after a follow up of 10 years.

Comment

In bone, xanthomatosis is rare and appears generally as a benign secondary manifestation of some diseases like: a...
nonossifying fibroma, fibrous dysplasia, anevrysmal bone cyst, a benign fibrous histiocytoma... The disease is nominated primary xanthoma when the other diseases are discarded. It is associated generally with hyperlipoproteinemia. In xanthoma with hyperlipoproteinemia, lipid deposits may occur on the trunk, the extremities and the bone marrow, which may be progressively infiltrated and replaced by lipid deposits, resulting in trabecular and cortical bone resorption. In xanthoma without lipid disorders, one of the Parker’s hypotheses was retained. He proposed the possible appearance of the lesion as a reactive response to intramedullary haemorrhage following a trauma. In our case report, the patient had a past history of trauma. Primary xanthoma in bones is mainly seen in men. The average age ranges from 20 to 50 years. In patients with lipid disorders, lesions are multifocal, located particularly in the lower limbs. The association with subcutaneous xanthomas is frequently observed. Whereas, in xanthoma with hyperlipoproteinemia, extra-osseous localizations are inexistent. Primary complaint is generally the pain. Symptoms are absent 30% of the time. Bone xanthomas are usually small. Radiologically, these tumors have a benign appearance with a purely lytic matrix and good margination. Surrounding sclerosis is frequent. The CT-scan and the MRI are useful to eliminate an extensive lesion. In costal xanthoma, differential diagnosis involve: a malignant tumor, fibrous dysplasia, nonossifying fibroma and cysts. The diagnosis is based on histologic findings. Diagnosis can be...
retained on a surgical biopsy or a needle biopsy specimen. Microscopically, foam cells, macrophages, cholesterol, fibrosis and hemosiderin are often associated. Histologic differential diagnosis includes Erdheim–Chester disease (a multisystemic lipid granulomatosis), Rosai–Dorfman disease and a metastatic clear cell carcinoma. If the diagnosis is difficult to establish, especially on a needle biopsy specimen, immunohistochemistry is very helpful. In xanthoma with lipid disorders, a medical treatment could be sufficient, whereas, in xanthoma without hyperlipoproteinemia, a complete or a partial removal of the lesion is effective. Berton et al. reported 21 cases of primary xanthoma without lipid disorders; the treatment approach was surgical in all cases in addition to a radiation therapy in three cases because of an incomplete curettage. After a follow up of 20 years, no recurrences were reported. The treatment of costal xanthomas is based on surgical removal. Surgical treatment is more necessary to exclude a malignant costal tumor with a benign radiologic findings than to cure the lesion by itself.

Conflict of Interest

None of the authors have a conflict of interest in relation to this work.

References