ORIGINAL ARTICLE

Post-traumatic fibro-osseous lesion of the ribs: a relatively under-recognised entity

S Aubert, O Kerdraon, M Conti, D Buob, S Petit, X Leroy

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See end of article for
authors' affiliations

Correspondence to:
S Aubert, Service
d'Anatomie Pathologique,
Pôle Biologie-Pathologie,
Parc Eurasanté, CHRU,
59045 Lille Cedex, France;
s-aubert@chru-lille.fr

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Aim: To report the description of a rare benign osseous lesion affecting the ribs entitled post-traumatic fibro-osseous lesion (PTFOL).

Methods: Seven cases of PTFOLs were retrieved from the archives of the University Hospital of Lille. Histological slides were reviewed and lesions were classified according to the histological patterns described by McDermott *et al.* Clinical and follow-up data were obtained from the patients' charts.

Results: PTFOLs occurred principally in men (mean age 31.8 years) with a known or suspected previous chest injury for four of them. No previous cancer was noted. Radiologically, PTFOLs readily presented as an isolated expansive lucency with a sclerotic rim located on the last five ribs. A constant increased uptake of radionuclide was noted on bone scan. Microscopically, two fibro-osseous, four xanthomatous and one mixed pattern were individualised. Lesions were characterised by a network of anastomosing bone trabeculae without osteoblast lining within a fibrous stroma. A zonal maturation from woven to peripheral lamellar bone was characterised. Central sheets of lipid-laden histiocytes were conspicuous in the xanthomatous type. For each patient, clinical follow-up was excellent, without any recurrence.

Conclusions: This is the second largest series of PTFOLs, which is considered to be a dysplastic healing process after trauma. It may be symptomatic or shown by imaging studies realised for unrelated reason. PTFOL is regularly misdiagnosed with other more common lesions of the ribs, such as fibro-osseous dysplasia, osteoma osteoid and benign fibrous histiocytoma. Its recognition is of importance because no follow-up is needed after resection.

The most common benign lesion of the rib is represented by fibrous dysplasia either in a monostotic or polyostotic manner.¹ Apart from fibrous dysplasia, other benign lesions such as osteoid osteoma, benign fibrous histiocytoma (BFH) and related illness have been described. Another relatively unknown diagnostic lesion that typically affects the ribs is represented by post-traumatic fibro-osseous lesion (PTFOL). This entity is thought to be a post-traumatic reactive process, which was clearly described by McDermott *et al.*² This study reports a series of seven cases of this entity.

PATIENTS AND MATERIALS

The computerised records of patients from 1991 to 2004 were searched for benign rib lesions surgically removed at the University Hospital of Lille, France. Two pathologists experienced in bone pathology (SA and XL) reviewed 124 cases and retrieved seven cases of PTFOL. Surgical material was fixed with formalin, decalcified with a hydrochloric acid-based solution, embedded with paraffin wax and stained with a solution of haematoxylin eosin saffron. Three to four slides were available for each case. Clinical and follow-up data were obtained from the patients' charts. When possible, patients were contacted to obtain additional clinical information.

RESULTS

Clinical data

Table 1 summarises the clinical data of the cases. Patients were predominantly male (6 men, 1 women) young adults (mean age 31.8 years, range 14–42 years). The initial presentation was post-traumatic for three patients at 2 (patient 2), 6 (patient 3) and 12 (patient 4) months, respectively, after a minor chest injury while practising sport (skiing, rugby and soccer, respectively). For one patient the lesion was incidentally found (patient 5), but he was

dependent on alcohol and had frequent falls (one prior nose fracture). Two patients presented with a painful rib (patient 1) and a costal neuralgia (patient 6), but did not recall any chest injury. No information was available for the last patient. None of them had a job with risk of chest injury and a history of cancer. No other bone lesion was found.

On radiological studies (roentgenogram, computed tomography or magnetic resonance imaging), lesions were found to be always unifocal and typically involved the last five ribs with a predilection for the posterior lateral rib (6/7 patients). They measured from 1–3.5 cm (mean 2.16 cm). Lesions were expansive in five patients with a central radiolucency and a sclerotic border (fig 1). Cortical rib was irregular (4/7 patients) with a disruption in three cases (fig 2). Bone scan, when available, always showed an increased uptake of radionuclide. Table 1 summarises the preoperative diagnoses. For patient 1, radiological findings were suggestive of a primary or secondary malignant process. For the other patients, despite the lack of evident sign of malignancy and because imaging findings were non-specific, a surgical resection was performed to rule out a tumour process.

In all patients, no sign of recurrence was observed at the last follow-up (range 12–145 months). The two patients who underwent the largest resection, however, presented local complications after 2 and 48 months, respectively. Indeed, patient 4 complained about persistent pain owing to an exudative pleural effusion that was treated by anti-inflammatory treatment and kinesitherapy. For patient 7, surgical resection was complicated by the apparition of a pulmonary hernia that was treated with a goretex mesh patch.

Abbreviations: BFH, benign fibrous histiocytoma; PTFOL, post-traumatic fibro-osseous lesion

Table 1 Clinical data of the seven cases of post-traumatic fibro-osseous lesion of the ribs

No	Age (years)	Sex	Trauma history	Delay (months)	Clinical presentation	Follow-up
1	36	M	No	—	Painful rib	Alive and well at 47 months
2	30	M	Sport (soccer)	2	Painful rib	Alive and well at 12 months
3	14	M	Sport (rugby)	6	Painful rib	Alive and well at 145 months
4	37	M	Sport (skiing)	12	Painful rib	Alive and well at 38 months
5	22	M	Frequent falls, previous nose fracture	—	Incidentally found	Alive and well at 95 months
6	42	F	No	—	Costal neuralgia	Alive and well at 72 months
7	42	M	N/A	—	N/A	Alive and well at 124 months

F, female; M, male; N/A, not applicable.

Pathological findings

Table 2 shows the pathological and radiological features of each case. Initially, three cases were diagnosed as a fibrous dysplasia, one case as a BFH, one case as an aneurysmal cyst and the last two recent ones as PTFOL. The five oldest cases were reclassified as PTFOL. Lesions were more or less centrally located and well demarcated from the adjacent marrow. According to Mc Dermott *et al*,² the histological pattern varied from fibro-osseous, xanthomatous to mixed.

The fibro-osseous pattern (2/7 cases) was characterised by a lace-like network of bone trabeculae with a zonal pattern of maturation within a bland fibroblastic stroma. No mitotic activity was noted. A metaplastic woven bone with irregular border was centrally located with mature peripheral lamellar bone. A clearly defined central stromal region containing a few isolated round to oval bone spicules was found, which melded with an increasing number of larger and anastomosing bone trabeculae, which eventually merged peripherally with the cortical bone. The stroma appeared oedematous, resembling that seen in nodular fasciitis, whereas in some areas it was more compact but without a whirling or storiform pattern. Typically, large blood vessels were seen at

the periphery. Rare fat cells were entrapped within the stroma. Osteoblasts were mainly absent, and whenever found were small and inconspicuous. Rare osteoclasts were present, where foam cells were seen. Sometimes, irregular fragments of lamellar bone were depicted within the fibro-osseous stroma (fig 3).

The xanthomatous variant pattern (4/7 cases) was characterised by a central, relatively large, sheet of histiocytes having pale foamy to granular cytoplasm within a loose fibrous stroma. Few small isolated metaplastic bone spicules were seen abutting a network of anastomosing bone trabeculae whose features were identical to those in the fibro-osseous cases, becoming continuous with the cortical bone (fig 4).

One patient had a mixed pattern, with a central region of anastomotic bone and aggregates of foam cells filling the intertrabecular spaces (fig 5).

DISCUSSION

Since the first description by Reed,³ <30 observations of PTFOL have been reported under various names in the literature: traumatic dysplasia, symmetrical fibro-osseous dysplasia of the rib; post-traumatic symmetrical fibro-osseous

Figure 1 Plain roentgenogram showing expansive lesion of the rib with a central radiolucency and a sclerotic border in the lateral seventh right rib (arrow).

Figure 2 Three-dimensional contrast enhanced scan showing expansive lesion with a dense border and focal cortical disruption.

Table 2 Pathological and radiological features of the seven cases of post-traumatic fibro-osseous lesion of the ribs

No	Size (cm)	Location	Roentgenogram	MRI or CT	Bone scan	Preoperative diagnosis	Initial pathological diagnosis	PTFOL histological subtype
1	3	Posterior-lateral 7 L	N/A	Expansive lesion with irregular cortex and cortical disruption	Positive	Malignant tumour, primitive or metastatic	PTFOL	Xanthomatous
2	N/A	Posterior-lateral 6 R	Well-defined lucency	Expansive lytic lesion	N/A	N/A	PTFOL	Xanthomatous
3	1.5	Anterior-lateral 7 R	Well-defined lucency	Lacunae with fracture	Positive	Eosinophil granuloma	BFH	Xanthomatous
4	1	Posterior-lateral 5 L	Expansive lesion	Lacunae with irregular cortex	Positive	Osteoid osteoma	Fibrous dysplasia	Osteofibrous
5	3.5	Posterior 6 L	Well-defined lucency	Well-defined mixed lytic/solid tumour with irregular cortex	N/A	Fibrous dysplasia	Aneurysmal cyst	Xanthomatous
6	2	Posterior 7 L	Expansive lesion	Expansive well-defined ovoid tumour	Positive	N/A	Fibrous dysplasia	Osteofibrous
7	2	Posterior-lateral 10 L	N/A	Expansive lytic lesion with cortical disruption	Positive	N/A	Fibrous dysplasia	Mixed

MRI, magnetic resonance imaging; CT, computed tomography; PTFOL, post-traumatic fibro-osseous lesions; L, left; R, right; N/A, not applicable; BFH, benign fibrous histiocytoma.

dysplasia, painless fibro-osseous lesion of the rib resembling osteoma osteoid; and sclerosing xanthoma.²⁻⁷ The true incidence of PTFOL is probably underestimated because it is regularly misdiagnosed as more common lesions of the ribs. PTFOLs are classically single rib lesions, but they may affect contiguous ribs, be polyostotic, or, more exceptionally, be solely extra costal, with one reported in the scapula by Kessler *et al.*⁷ PTFOL is systematically reported to occur on the five last ribs, as signalled in the present work.

The series of McDermott *et al.*² reported 11 lesions of the ribs in patients with a mean age of 54.6 years (range 31–79 years). A traumatic history was found in seven patients. Delay between traumatic episode and the discovery of the lesion could be variable, ranging from 2 to 42 years. Ideally, the three patterns of PTFOL could have represented the successive stages of the same lesion. But, as shown by McDermott *et al.*,² there is no temporal relationship between the episode of trauma and the histological pattern. The chest trauma is often described as minor and subclinical, and so it is sometimes unrecognised owing to being common to the ribs. PTFOLs are usually described as incidental findings, but they may be symptomatic, discovered during investigations of persistent chronic localised pain related to the lesion, as seen in most of our patients. Radiologically, roentgenogram may be negative and the lesion may only be discovered as a

hot spot on bone scan. Elsewhere, a PTFOL typically appears as a more or less expansive or fusiform central radiolucency with a sclerotic rim, whereas some of them have been reported as radiodense.^{2 4 5 8}

In McDermott series,² during investigation 6 of the 11 patients were discovered to have a concomitant cancer (mainly prostatic carcinoma). This raised the question of an eventual metastatic unique location of a primitive cancer in the differential diagnosis, although this is a rare event on the ribs.² The series in this study differed by a younger age at presentation and the absence of cancer history. So, a monostotic form of fibrous dysplasia represented the main microscopic differential diagnosis, in particular with the fibro-osseous pattern of PTFOL. Indeed, three patients were initially diagnosed with a fibrous dysplasia. Fibrous dysplasia is the most frequent benign lesion of the ribs. Importantly, however, this benign lesion of the ribs contrasts with the random appearance of the woven bone that is set in a delicate fibrous stroma. Trabeculae are thinner, more irregularly shaped, without the woven to lamellar bone maturation. Xanthomatous cells, when present, are rarely in a sheet-like manner. This distinction is of importance because the diagnosis of fibrous dysplasia implies radiological exploration and a long clinical follow-up with the risk, although minimal, of a sarcomatous transformation.¹

Figure 3 Fibro-osseous pattern presenting anastomosing bone trabeculae with zonal maturation within a fibrous stroma (magnification $\times 2.5$).

Figure 4 Xanthomatous pattern with central area of foam cells within a loose fibrous and highly vascular stroma (magnification $\times 5$).

Figure 5 Mixed pattern with maturing bone trabeculae separated by aggregates of foam cells (magnification $\times 10$).

Another important diagnosis to be considered was that of osteoid osteoma. Indeed, the combination of trabeculae anastomoses and sclerotic rim in PTFOL could be suggestive of osteoid osteoma.⁵ Osteoid osteoma is uncommon in patients over 40 years and rarely occurs in the ribs, but when it does it is virtually always accompanied by severe pain.⁹ Microscopically, osteoid osteoma differed from PTFOL by a greater degree of stroma vascularity, osteoid and bone trabeculae, typically randomly distributed without a zonal pattern and most significantly lined by numerous plump osteoblasts.

BFH should also be discussed in the differential diagnosis. Indeed, ribs and the ilium are the most commonly affected sites. Radiologically, it typically appears as a well-demarcated radiolucent medullary defect with a sclerotic rim. BFH is microscopically indistinguishable from non-ossifying fibroma, and the confusion with the xanthomatous variant of PTFOL is at most when lipid-laden histiocytes are numerous (so-called xanthoma, xanthofibroma). BFH, however, is classically painful and microscopically presents a whorled fibrous stroma punctuated by giant cells and scattered inflammatory cells. Reactive osteogenesis lined by plump osteoblasts may be present at the periphery, but it lacks the woven to lamellar maturation with a zonal pattern as seen in PTFOL.^{10 11}

The xanthomatous variant of PTFOL may also be confused with other lipid-filled lesions such as Erdheim–Chester disease and eosinophilic granuloma.

Erdheim–Chester disease affects multiple bones that show an increased cortical bone density. The ribs are rarely implicated and are usually mentioned as a part of multiple skeletal involvement. Microscopically, there is a diffuse lipid-laden histiocyte infiltration with lymphocytes, plasma cells and occasional eosinophils filling the marrow spaces. It is associated with irregular bone thickening with a variable number of osteoclasts and osteoblasts.^{12 13}

A regressive stage of eosinophilic granuloma, although rare in adults, may affect the ribs. On imaging study, it presents as a sharply demarcated lucency surrounded by a sclerotic

border. Microscopically, regressing eosinophilic granuloma shows massive fibrosis and the surrounding bone is reactive without the anastomosing and zonal pattern of PTFOL.¹¹ If a doubt persists, an immunohistochemical study with the antibody directed against CD1a may help to correct the diagnosis in favour of an eosinophilic granuloma.

In some aspects, PTFOL resembles hypertrophic callus after trauma. Indeed, fracture callus shows a gradual transition from hypercellular areas containing spindle cells with early osteoid formation of an interconnecting network of reactive bone. More mature lesions show progressive maturation to lamellar bone. Unlike PTFOL, however, bone trabeculae are rimmed by numerous plump osteoblasts and osteoclasts. For some investigators, PTFOL could correspond to an abnormal osseous healing process, leading to its incomplete maturation. Their exclusive occurrence on flat bones would be linked to insufficient mechanical constraints to enable maturation in lamellar bone, inducing an abortive form of healing.^{3 4 6}

Because no recurrence has ever been reported, no follow-up is needed after limited resection. In accordance with other authors, it seems reasonable to propose a simple radiological surveillance of patients presenting solitary and asymptomatic rib lesion, especially in the context of a prior chest injury.^{2 5}

In conclusion, this study is the second largest series in the literature. PTFOL is a benign osseous lesion after a subclinical trauma that occurs mainly to the ribs. This under-recognised entity should be studied to avoid frequent misdiagnosis with more common lesions in this location. Moreover, no follow-up is needed after limited resection.

Authors' affiliations

S Aubert, O Kerdraon, D Buob, S Petit, X Leroy, Department of Pathology, University Hospital, CHRU, Lille, France
M Conti, Department of Thoracic Surgery, Calmette Hospital, CHRU

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