Case Report

J Korean Soc Radiol 2022;83(2):414-419
https://doi.org/10.3348/jksr.2021.0108
eISSN 2288-2928

대한영상의학회지

Florid Reactive Periostitis of the Clavicle: A Case Report

쇄골에 발생한 개화성 반응성 골막염: 증례 보고

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Received June 11, 2021
Revised August 11, 2021
Accepted October 18, 2021

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Florid reactive periostitis (FRP) is a rare benign fibro-osseous proliferation, occurring mostly in the short tubular bones of hands and rarely in the long tubular bones. We report a surgically confirmed case of FRP involving the clavicle in a 26-year-old male. On MRI scans, a soft tissue mass with T2 high signal intensity was found that originated from the periosteum of the clavicle and included surrounding a periosteal elevation and perilesional soft tissue edema. Strong contrast enhancement was noted inside the mass and along the periosteum involving more than half of the circumference of the clavicle. Serial radiographs revealed a soft tissue mass without mineralization that turned into an ossified mass with a solid periosteal reaction within a month.

Index terms Periostitis; Fasciitis; Clavicle; Neoplasms

INTRODUCTION

Florid reactive periostitis (FRP) is a rare benign fibro-osseous proliferation, reported mostly in the short tubular bones of hands and rarely in the long bones (1, 2). To the best of our knowledge, this is the first case of FRP involving the clavicle that has been reported in the English literature to date. Herein, we report a case of FRP involving the clavicle with radiographic and MRI findings.

CASE REPORT

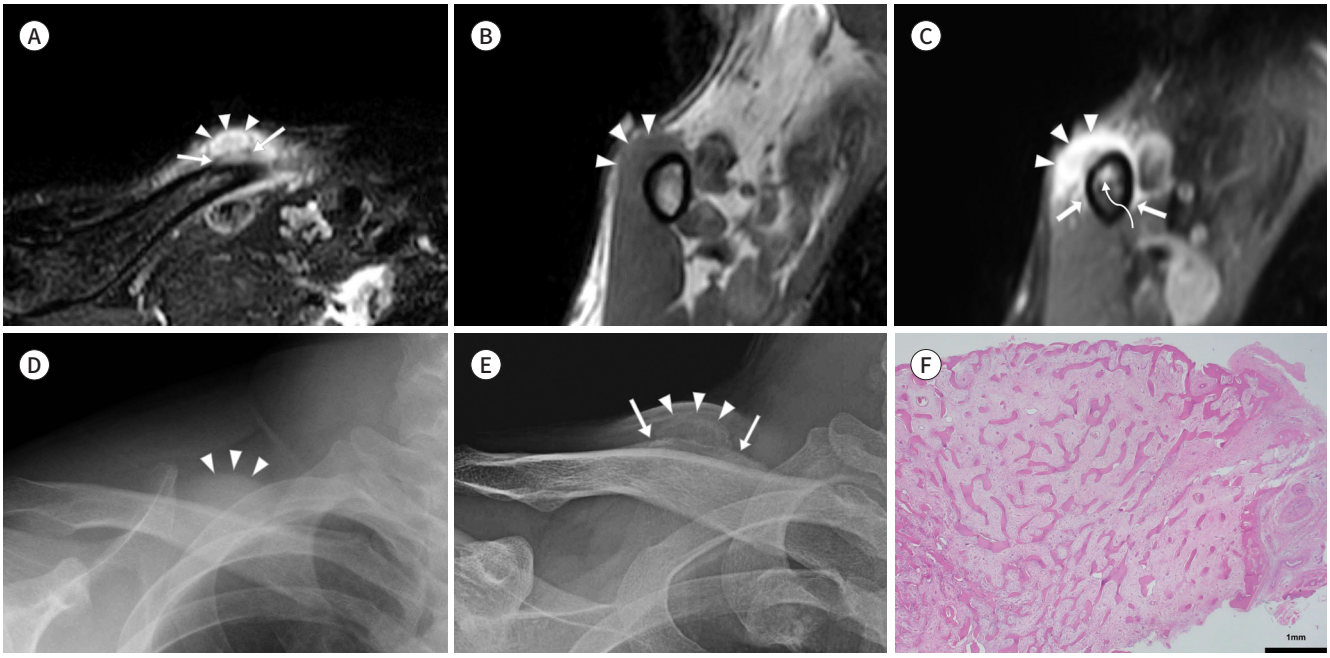
A 26-year-old male visited with a painful soft tissue mass at his right clavicle. He denied any history of trauma. He had noticed the mass 2 weeks ago and the size of the mass had been increasing with worsening of pain. The clinical impression was osteomyelitis because of the rapidly growing nature of the mass with tenderness, but all lab-

oratory findings were within normal limits.

MRI was obtained 3 days after the initial visit and showed a small mass with T2 high signal intensity abutting the midshaft of the clavicle. Periosteal elevation was noted adjacent to the origin of the mass (Fig. 1A). The mass had iso signal intensity to the adjacent pectoralis major muscle on T1 weighted image. The cortex of the clavicle was relatively preserved without disruption and normal high signal intensity was maintained in the bone marrow on T1 weighted image (Fig. 1B), which allows the exclusion of a malignant disease infiltrating the bone marrow. The mass revealed strong contrast enhancement after contrast administration and also did the periosteum involving more than half of the circumference of the clavicle midshaft. And focal edema was noted in the abutting bone marrow (Fig. 1C). From these MR findings, the initial imaging diagnosis was a thrombosed intracortical hemangioma. Clinical follow-up was scheduled after 2 weeks to see whether there is a change in size or character of the mass within this period.

As the mass did not show any change in 2 weeks, surgical excision was planned and the

Fig. 1. Imaging findings in a 26-year-old male revealing florid reactive periostitis involving the clavicle.
A. MR T2 weighted axial image shows a well-defined ovoid-shaped mass (arrowheads) with T2 high signal intensity arising from the periosteum of the clavicle with an adjacent periosteal elevation (arrows).
B, C. On MR pre- and post-contrast T1 weighted sagittal images, the mass (arrowheads) shows iso-signal intensity compared to the adjacent pectoralis major muscle on pre-contrast images and shows strong contrast enhancement after contrast-agent administration. Normal high signal intensity of the bone marrow was maintained in the medullary cavity on T1 weighted image with preserved cortex, although focal bone marrow edema was observed (curved arrow, C). Note the contrast enhancement along the periosteum (arrows, C) covering more than half of the circumference of the involved clavicle.
D. Chest radiograph, two weeks after MR, shows a radio-dense soft tissue mass (arrowheads) without definite mineralization at the right supraclavicular area.
E. On a pre-operative radiograph, six weeks after MR, the mass (arrowheads) shows rapid ossification within four weeks with solid periosteal reaction (arrows) at the right mid clavicle.
F. Histopathology. The mass is composed of woven bone rimmed by osteoblasts with proliferation of cytologically bland spindle cells (hematoxylin and eosin stain, × 12.5).



preoperative anteroposterior chest radiograph was obtained at this time point. The mass did not show any mineralization on the chest radiograph (Fig. 1D).

However, the mass rapidly became firm and hard within several weeks before the surgery. And preoperative radiograph of the clavicle, which was taken 6 weeks after MR, revealed peripherally predominant ossification of the mass with adjacent solid continuous periosteal reaction (Fig. 1E).

The patient underwent surgical excision. The mass was easily dissected from the closely abutting clavicle and also from the pectoralis major muscle. Histopathology revealed spindle cell proliferation with immature bone formation rimmed by uniform osteoblasts (Fig. 1F). Inflammatory cell infiltration was not observed, and there were no atypical mitotic figures nor pleomorphism suggesting malignancy. Microscopically, this lesion was similar to the myositis ossificans, but the zoning pattern was poorly detected. These features were considered diagnostic of FRP.

This report was approved by the Institutional Review Board and the requirement for informed consent was waived (IRB No. 20-2021-3).

DISCUSSION

Histologically, FRP shows reactive proliferation of fibro-osseous components, which is a common feature of benign fibro-osseous lesions such as myositis ossificans, fasciitis ossificans, and bizarre parosteal osteochondromatous proliferation. Nevertheless, they have subtle differences in involved location and microscopic findings. Myositis ossificans is a reactive bone-forming lesion with the proliferation of spindle-shaped fibroblasts, most commonly occurs within the muscles and is characterized by the typical zoning pattern (inner cellular zone resembling nodular fasciitis with spindle cell proliferation; intermediate zone with woven bone formation; outer zone with mature lamellar bone). Similar lesions in the tendons or fascia are known as fasciitis ossificans, whereas similar lesions involving the periosteum are considered FRP. However, Bizarre parosteal osteochondromatous proliferation is composed of aggregated cartilage, new bone, and fibrous tissue. In this patient, myositis ossificans was excluded because the mass was not originated from the muscle and Bizarre parosteal osteochondromatous proliferation was also excluded due to the lack of cartilage tissue. Meanwhile, Yamamoto et al. (3) reported a case of parosteal fasciitis involving the clavicle with emphasis on the pathologic findings, in which the MR image seemed very similar to our presented case. However, nodular or parosteal fasciitis is mainly composed of proliferated plump fibroblastic/myofibroblastic cells whereas osseous metaplasia is only seen occasionally (4). Therefore, we considered FRP as the best diagnosis of the presented case, based on the combination of clinical course, radiologic and pathologic findings.

Clinical presentation of FRP is usually a soft tissue swelling, sometimes with progressively worsening pain within several weeks or months (5). History of trauma was reported in less than half of the cases (1, 2). Despite many cases reported to be spontaneously regressed without any treatment (1, 2), the lesion is often excised to exclude the possibility of malignancy.

The imaging findings of FRP involving the long tubular bone are not different from those involving the short tubular bone (2). Radiographic findings of FRP are soft tissue mass adja-

cent to a bone showing a variable degree of calcifications/ossifications and periosteal reaction (6). Cortical irregularity and erosion, which raises the concern about the malignant lesion, are not uncommon (2, 5). Above all, rapid periosteal bone production without change of soft tissue mass may be the most characteristic finding of FRP (7). In our reported case, the mass was mainly soft tissue mass without mineralization until 4 weeks after the presentation. But, the mass showed rapidly progressive ossification and adjacent solid periosteal reaction after another 4 weeks and this radiographic change was the most diagnostic feature of FRP. Peripherally predominant ossification, a semi-zoning pattern, was described in the largest case series of FRP involving the long tubular bones by Jamshidi et al. (2). The "zoning pattern" was formerly known as a typical pathologic finding of myositis ossificans, but recent investigation has noted that FRP can show a similar semi-zoning pattern (in 2 out of 7 cases), which is formed by immature spindle cells and woven bones in the center along with the mature bone at the periphery. Because this semi-zoning pattern can be the differentiating feature of FRP from the malignant tumors, the intralesional ossification pattern should be carefully evaluated for a juxtacortical soft tissue mass abutting the long bones. Likewise, MR imaging should always be interpreted in conjunction with plain radiographs not to miss such an important clue.

MR findings of FRP were mostly soft tissue masses adjacent to the shaft of long tubular bones with surrounding soft tissue edema, frequently accompanied by cortical erosion and marrow edema in the abutting bone (2). The periosteal reaction was almost always present, but the amount of new bone formation varied. MRI of our reported case showed consistent findings; a juxtacortical mass with T2 high signal intensity arising from the midshaft periosteum of the clavicle, a long tubular bone, and adjacent periosteal elevation. The periosteum was the most suspected origin of the mass, considering its wide attachment at the clavicle, periosteal elevation adjacent to the mass, and circumferential enhancement along the periosteum around the clavicle.

The interesting clinical course of FRP involving the long tubular bones, reported by Jamshidi et al. (2), was spontaneous regression of all lesions with a variable degree of residual exostosis which was formed by a cortical thickening without a corresponding medullary cavity. Therefore, when FRP involving a long bone can be properly diagnosed on imaging, unnecessary surgery can be avoided. In the reported case, the surgery was planned because the MR and radiographic studies were done in a very early phase of the disease and did not show the typical ossification pattern on the radiographs. Therefore, short-term follow-up radiographs, CT, or US can be helpful to notice the typical ossification pattern for the correct diagnosis.

Radiologic differential diagnosis of FRP involving the long tubular bone includes various surface lesions of the bone such as intracortical abscess, osteoid osteoma, subperiosteal hematoma, and malignant surface tumors of the bone. The intracortical abscess may show a similar appearance to FRP on MR, however, minimal involvement of the cortex or medullary cavity and lack of laboratory abnormality can help to exclude infectious conditions (2, 8). Osteoid osteoma can involve the outer cortex and show extensive periosteal reaction with surrounding reactive changes on MR. But lack of round osteolytic nidus, which would be more evident on CT, can help to differentiate FRP from an osteoid osteoma. Subperiosteal hematoma can be ossified only after a long period to demonstrate similar radiographic findings of

FRP. In addition, an ossified subperiosteal hematoma usually does not show contrast enhancement on MR nor reactive change around the lesion (9). It can be very difficult to distinguish early FRP from a small malignant lesion in the bone surface. However, follow-up radiographs or CT can demonstrate rapid maturation of periosteal reaction into a solid ossification within several months in FRP (7, 10), which is quite different from irregular mineralization or interrupted periosteal reactions of malignant surface tumors. The biopsy can be recommended for confusing cases to exclude malignancy.

Here, we report a case of FRP involving the clavicle, which is the first case in the English literature. The key clinical and radiologic findings of FRP were a painful growing soft tissue mass abutting the outer cortex of a long tubular bone midshaft without involving the medullary cavity, strong contrast enhancement of the mass with the adjacent linear periosteal reaction on MR, and finally, the rapid subsequent ossification of the mass with semi-zoning pattern and adjacent solid periosteal reaction on radiographs. The radiologists should consider FRP when this typical clinical course and imaging findings are present.

Author Contributions

Conceptualization, C.J.W., J.C.H.; formal analysis, C.J.W., P.H.E.; investigation, K.D.H., K.H.J., S.J.; writing—original draft, all authors; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Funding

None

REFERENCES

1. Spjut HJ, Dorfman HD. Florid reactive periostitis of the tubular bones of the hands and feet. A benign lesion which may simulate osteosarcoma. *Am J Surg Pathol* 1981;5:423-433
2. Jamshidi K, Givehchian B, Mirzaei A. Florid reactive periostitis of the long bone: a case series of seven patients. *J Orthop Sci* 2017;22:560-565
3. Yamamoto T, Marui T, Mizuno K, Obayashi C, Minami R, Inaba M, et al. Parosteal fasciitis of the clavicle. *Pathol Int* 2000;50:987-991
4. Kwittken J, Branche M. Fasciitis ossificans. *Am J Clin Pathol* 1969;51:251-255
5. Howard RF, Slawski DP, Gilula LA. Florid reactive periostitis of the digit with cortical erosion: a case report and review of the literature. *J Hand Surg Am* 1996;21:501-505
6. Jambhekar NA, Desai SS, Puri A, Agarwal M. Florid reactive periostitis of the hands. *Skeletal Radiol* 2004; 33:663-665
7. Sundaram M, Wang L, Rotman M, Howard R, Saboeiro AP. Florid reactive periostitis and bizarre parosteal osteochondromatous proliferation: pre-biopsy imaging evolution, treatment and outcome. *Skeletal Radiol* 2001;30:192-198
8. Gao Z, Wang J, Wang Z, Meng Q. Florid reactive periostitis of the metacarpal and phalanx: 2 case reports. *J Hand Surg Am* 2013;38:2134-2137
9. Guillin R, Moser T, Koob M, Khoury V, Chapuis M, Ropars M, et al. Subperiosteal hematoma of the iliac bone: imaging features of acute and chronic stages with emphasis on pathophysiology. *Skeletal Radiol* 2012; 41:667-675
10. Seeger LL, Yao L, Eckardt JJ. Surface lesions of bone. *Radiology* 1998;206:17-33

쇄골에 발생한 개화성 반응성 골막염: 증례 보고

박혜은[1,2] · 채지원[3,4]* · 조현철[5,6] · 김지은[1,2] · 김동현[3,4] · 김효진[3,4] · 서지운[3,4]

개화성 반응성 골막염은 드문 양성 섬유골성 증식으로 대부분 손의 짧은 관상골을 침범하고, 긴 관상골의 침범은 드물다. 저자들은 26세 남자에서 수술로 확진되었던 쇄골의 개화성 반응성 골막염 1예를 보고한다. 자기공명영상에서 병변은 높은 T2 신호강도를 보이는 연조직 종괴로 쇄골의 골막에서 기인하는 것으로 보였고, 주변의 골막거상과 종괴 주변의 연조직 부종이 관찰되었다. 조영증강 시 종괴의 내부는 강한 조영증강을 보였고, 쇄골의 골막을 따라서도 쇄골 둘레의 절반 이상을 둘러싸며 조영증강이 확인되었다. 연속된 단순촬영에서는 무기질 침착이 없는 연조직 종괴로 보이다가 한 달 이내에 빠른 속도로 연조직 종괴의 골화가 확인되었고, 고체형 골막반응이 동반되었다.

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