ISSN: 2233-601X (Print) ISSN: 2093-6516 (Online)

http://dx.doi.org/10.5090/kjtcs.2013.46.6.478

□ Case Report □

Nonossifying Fibroma of the Rib Resected by Video-Assisted Thoracoscopic Surgery with Preservation of Periosteum

Ju Yeon Pyo, M.D.¹, Soon-Ho Chon, M.D., Ph.D.², Jae Yoon Ro, M.D., Ph.D.³

Nonossifying fibromas are not uncommon, but those described in the rib are unique. We report the case of a 15-year-old patient with symptoms of chest wall pain for 5 days who underwent a video-assisted thoracoscopic rib resection for a 2.5-cm rib mass. Unexpectedly, pathological results revealed a nonossifying fibroma of the rib. The results showed excellent cosmesis and new bone formation because of the preservation of the overlying periosteum.

Key words: 1. Benign tumor

- 2. Ribs
- 3. Video-assisted thoracoscopic surgery (VATS)

CASE REPORT

A 15-year-old soccer player visited our thoracic and cardiovascular surgery department with symptoms of right chest wall pain 5 days after the onset of the pain; the symptoms were experienced after a case of blunt trauma while playing soccer. The rib radiographic series revealed a 2.5-cm cystic lesion in the medullary portion of the posterior portion of his sixth rib, and a computed tomographic scan revealed an expansile cystic mass (Fig. 1). A technetium 99m (99mTc) methylene diphosphonate bone scan showed increased uptake. Surgery was recommended because of the uncertainty of the diagnosis. One month later, a rib resection by video-assisted thoracoscopic surgery was performed.

The patient was placed in a lateral position with three

ports: a camera port in the sixth intercostal space in his mid-clavicular line, a 3-mm port above his areola posterior to the camera port close to his mid-clavicular line, and a third 12-mm port in his areola margin. The tumor was identified and cut with 2-cm margins on both sides. Electrocautery and a periosteal elevator were used for preserving the overlying periosteum, and the rib was cut with a bone punch. The rib was removed through the areolar port and sent for a pathological examination.

The resected rib revealed a focal fusiform expanded lesion by an intramedullary well demarcated, grayish tan, solid mass with a lobulated margin, measuring 2.2×1.5 cm (Fig. 2). Microscopically, the mass was composed of spindle cells arranged in a storiform and interlacing fascicular pattern with foci of multinucleated giant cells and foamy histiocytes (Fig.

¹Department of Pathology, Hanyang University Guri Hospital, Hanyang University College of Medicine, ²Department of Thoracic and Cardiovascular Surgery, S-Jungang Hospital, ³Department of Pathology and Genomic Medicine, The Methodist Hospital, Weill Medical College of Cornell University

[†]This article was presented at the International Society for Minimally Invasive Cardiothoracic Surgery May 30th to June 2nd, 2012 in Los Angeles as an e-poster presentation.

Received: April 9, 2013, Revised: May 30, 2013, Accepted: June 3, 2013

Corresponding author: Soon-Ho Chon, Department of Thoracic and Cardiovascular Surgery, S-Jungang Hospital, 91 Wollang-ro, Jeju 690-232, Korea

⁽Tel) 82-64-786-7572 (Fax) 82-64-786-7999 (E-mail) sh.chon@hotmail.com

[©] The Korean Society for Thoracic and Cardiovascular Surgery. 2013. All right reserved.

[©] This is an open access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (http://creative-commons.org/licenses/by-nc/3.0) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.



Fig. 1. A 2.5-cm expansile mass (arrow) seen in the patient's sixth rib.

3A, B, C). Focal fracture callus was noted in the peripheral region around the cortical bone, characterized by the formation of hyperhplastic cartilaginous tissue and mature bony tissue. Immunohistochemical staining showed a positive reaction for smooth muscle actin in the spindle cells (Fig. 3D).

The patient's postoperative course has been uneventful thus far. His drain was removed on the second postoperative day, and he was discharged the day after. There are no signs of recurrence 1 year after the operation, and he is without pain and more than happy with the cosmesis. Postoperative 1-year chest computed tomographic scans have shown new normal bone formation in place of the removed benign rib tumor.

DISCUSSION

Nonossifying fibroma (NOF) was first described in 1942 by Jaffe and Lichtenstein [1] as a benign entity of bone, with spindle-shaped cells interspersed with multinucleated giant cells and foamy histiocytes. Since then, there have been numerous reports of similar cases described with terms such as fibrous cortical defect, nonosteogenic fibroma, xanthic variant of xanthoma, xanthogranuloma of bone, and metaphyseal fibrous defect [2]. Nonossifying fibromas of the rib are extremely rare. They are benign lesions characterized by storiform interlacing bundles composed of spindle-shaped fibroblasts, multinucleated giant cells, scattered lymphocytes, and foamy histiocytes [3]. The term 'nonossifying fibroma' trig-



Fig. 2. Gross specimen reveals a fusiform-shaped intramedullary lesion. Each unit represents 5 mm.

gers considerable confusion. It may be confused with fibrous cortical defect, which has an identical histology, but can be distinguished by its smaller size (less than 2 cm) [4] and its location in the metaphysic of long bones. The histological features of benign fibrous histiocytoma are similar to those of NOF. The term 'benign fibrous histiocytoma' is applied only if the lesion occurs in an unusual location in adults, while NOFs occur almost exclusively in the first two decades of life. NOFs are very commonly seen in the metaphysis of femur, tibia, and fibula of children but are rarely seen elsewhere. NOFs are known to occur eccentrically but can occur over the entire width of the bone [5]. NOFs in other locations have only been reported in the mandible, only nine of which have been reported in the literature [3]. Further, there are few known reports of NOFs occurring in the rib.

The radiographic findings of NOF have been established. In simple X-rays, they appear as a radiolucent lesion with a thinned cortex and sclerotic margin. In computed tomographic scans, NOFs may appear as a lesion with central radiolucency. Magnetic resonance imaging may reveal variable signal intensities with septations, more often hypointense on T1-weighted images. Mapping with fluorodeoxyglucose positron emission tomography scans may also be helpful with standardized uptake value of less than 2.5. Anything higher would exclude the diagnosis of NOF and would potentially indicate a more aggressive type of tumor such as osteosarcoma, chondrosarcoma, Ewing's sarcoma, or malignant lymphoma [6].

Nonossifying fibromas have been claimed to be associated

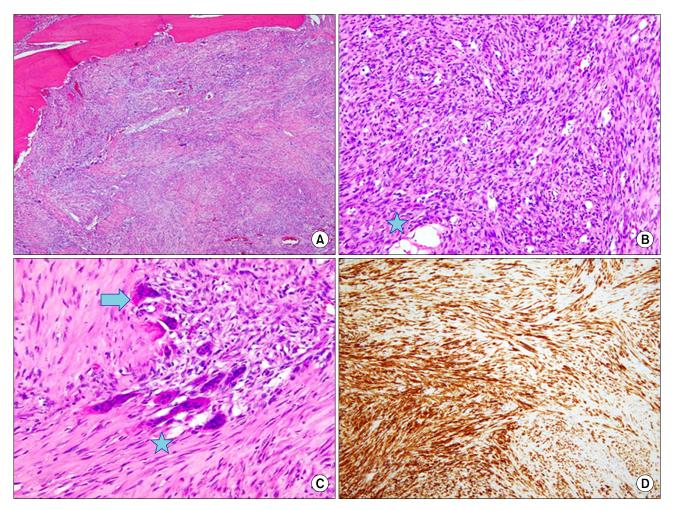


Fig. 3. (A, B, C) Spindle cells are arranged in storiform and interlacing fascicular patterns with foci of multinucleated giant cells (arrow) and foamy histiocytes (H&E, ×40, ×200, and ×400, respectively) (star). (D) Immunostaining for smooth muscle actin shows a diffuse positive reaction in spindle cells (×400).

with trauma as was also seen in our case [1]. Our case also displayed evidence of a fracture line presumably caused by the thinning of the patient's cortex. A nonossifying fibroma is considered to be a self-limiting entity that disappears by 29 to 53 months [3]. In our case, because of the uncertainty of the diagnosis, a surgical biopsy was inevitable. If the lesion were suspected despite its unusual location, an operation could have been prevented. We believe that the minimally invasive nature of the operation instead of a large incision that would be required in an open procedure may have provided better cosmetic outcome and less pain.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article has been reported.

REFERENCES

- 1. Jaffe HL, Lichtenstein L. *Non-osteogenic fibroma of bone*. Am J Pathol 1942;18:205-21.
- 2. Nelson M, Perry D, Ginsburg G, Sanger WG, Neff JR, Bridge JA. *Translocation* (1;4)(p31;q34) in nonossifying fibroma. Cancer Genet Cytogenet 2003;142:142-4.
- 3. Abdelsayed RA, Sharma S, Ferguson H. Fibrous cortical defect (nonossifying fibroma) of the mandibular ramus: report

- $\it of~2~cases.$ Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;110:504-8.
- 4. Dormans JP, Pill SG. Fractures through bone cysts: unicameral bone cysts, aneurysmal bone cysts, fibrous cortical defects, and nonossifying fibromas. Instr Course Lect 2002; 51:457-67.
- Unni KK, Inwards CY, Bridge JA, Kingblom LG, Wold LE. Tumors of the bones and joints. Washington (DC): American Registry of Pathology Armed Forces Institute of Pathology; 2005
- 6. Hetts SW, Hilchey SD, Wilson R, Franc B. Case 110: Nonossifying fibroma. Radiology 2007;243:288-92.