CASE REPORT

Osteoid osteoma of the lung: an extremely rare case report

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ABSTRACT

Osteoid osteoma was first described by Jaffe in 1935 and accounts for 10%–12% of all benign bone tumors. It is not aggressive nor has the potential for malignant transformation. This bone tumor often affects the long bones of the femur and tibia. Osteoid osteoma is small, osteoblastic, well-demarcated, benign bone tumor with a typical size of <1 cm and a characteristic surrounding zone of reactive bone. Extraskeletal osteoid osteoma of the lung is extremely rare. In the lung, there are tumors with an osseous element, such as hamartoma and amyloid tumor, and some extremely rare reactive lesions such as osseous metaplasia. The present work describes an extremely rare case of osseous osteoma of the lung. Studying the literature, we found that this is only the second case of osteoid osteoma of the pulmonary parenchyma.

Keywords: osteoid osteoma, lung

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INTRODUCTION

Osteoid osteoma was first described by Jaffe in 1935 [1]. Osteoid osteoma is a rare tumor that accounts for only 3% of primary bone tumors. People between the ages of 5 and 24 are most often affected. The main symptom of the disease is pain that worsens during the night and is relieved by aspirin [2]. Osteoid osteoma is small, osteoblastic, well-demarcated, benign bone tumor with a typical size of <1 cm and a characteristic surrounding zone of reactive bone [3]. The tumor can occur anywhere in the cortex or marrow of the bone. However, damage usually affects the long bones of the lower limbs [4,5]. Osteoid osteoma also occurs in the spine and flanks and is sometimes associated with scoliosis [6,7]. Extraskeletal osteoid osteoma of the lung is extremely rare. In the lung, there are tumors with an osseous element, such as hamartoma and amyloid tumor, and some extremely rare reactive lesions such as osseous metaplasia [8]. The present communication describes an

extremely rare case of osseous osteoma of the lung.

CASE PRESENTATION

A 77-year-old woman comes from a provincial hospital complaining of a productive cough with accompanying brownish/yellowish sputum since two months. From the individual medical history chronic atrial fibrillation under acenocoumarol, gastritis, gastroesophageal reflux, two normal deliveries, depression since one year, appendicectomy, tonsillectomy, and open cholecystectomy are reported. A paraclinical examination was performed with chest computed tomography and PET-CT, from which a nodular shadowing (23x18 mm) of the posterior bronchopulmonary part of the right upper lung lobe (SUVmax :3.2) was performed. (Figures 1-3). Following this, a wedge resection of the pulmonary lesion was performed (Figure 4).

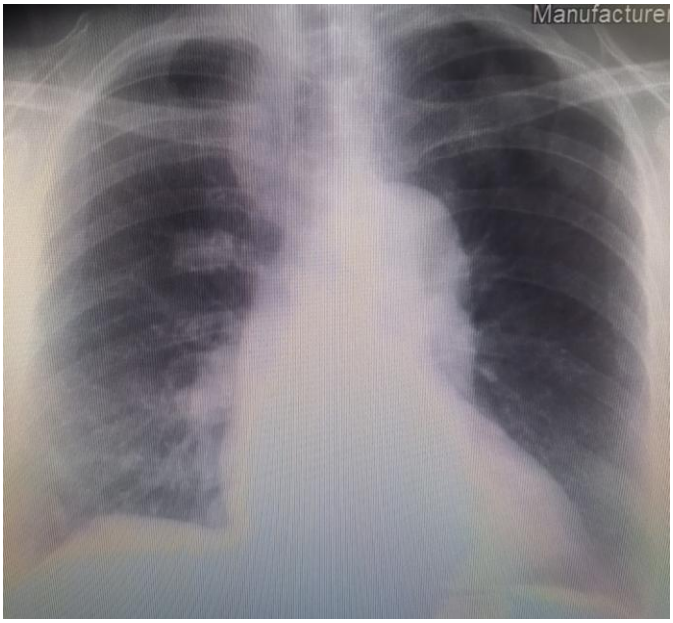

Figure 1. Preoperative chest X-ray. Right lung nodule with a smooth outline.

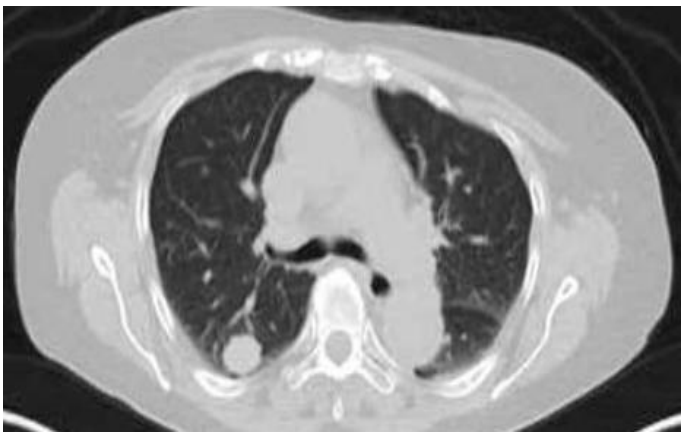

Figure 2. Preoperative computed tomography of the chest. Right upper lobe nodule with a smooth outline.

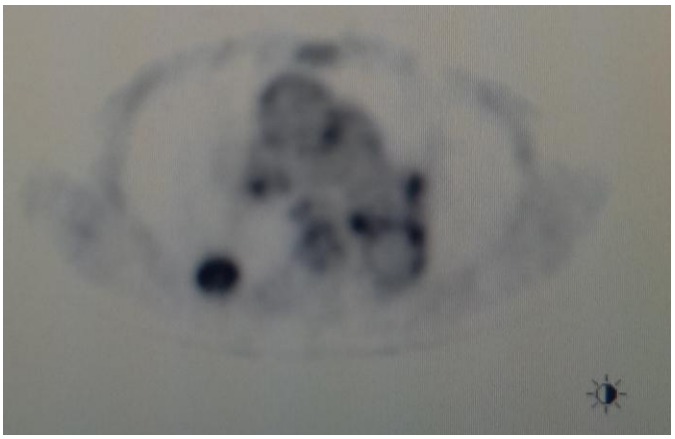

Figure 3. PET-CT. A nodular shadowing (23x18 mm) of the posterior bronchopulmonary part of the right upper lung lobe with (SUVmax :3.2).

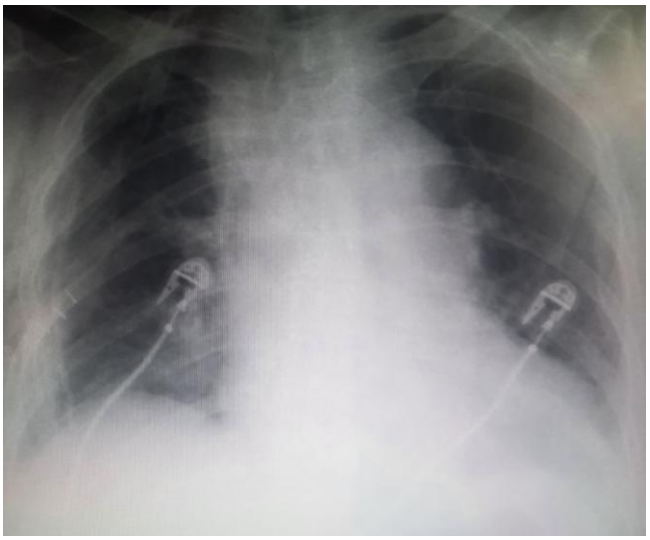

Figure 4. Postoperative chest x-ray.

A rapid biopsy proves that it is a benign tumor. Definitive histological evaluation demonstrates morphologic and immunophenotypic findings of osseous osteoma of the lung (Figures 5,6).

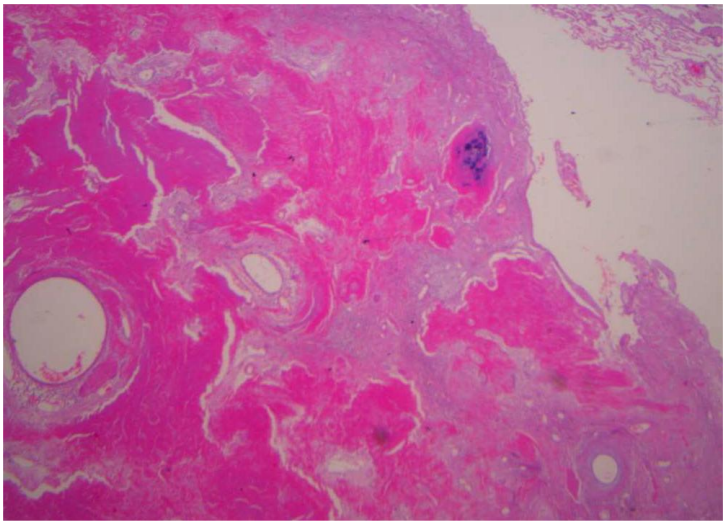

Figure 5. Eosin-eosinophyllin staining X25

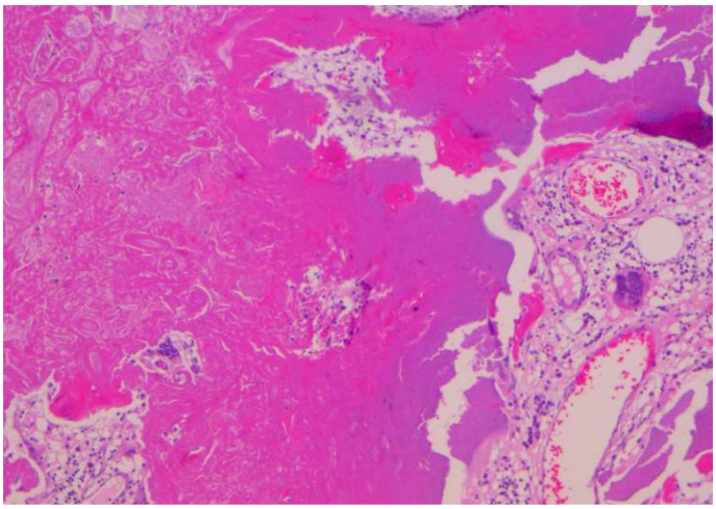

Figure 6. Eosin-eosinophyllin staining X100

COMMENT

Osteoid osteoma was first described by Jaffe in 1935 and accounts for 10%–12% of all benign bone tumors. [9] It is not aggressive nor has the potential for malignant transformation. This bone tumor often affects the long bones of the femur and tibia. The foot is less commonly affected (2-10%) with the ankle being most commonly affected [10]. An osteoid osteoma is usually surrounded by sclerotic bone. Osteoid osteomas do not exceed 2 cm in diameter and are classified into cortical, cancellous, and subperiosteal subtypes.

Osteoid osteomas occurring in long bones are mainly intracortical. The majority of osteoid osteomas present in the foot show minimal periosteal reaction. Size is used to differentiate an osteoid osteoma from an osteoblastoma. Osteoblastomas are usually larger than 2 cm while osteoid osteomas are less than 1.5 cm [11].

In the Bibliography various theories are developed regarding the pathophysiology of osteoid osteomas. One theory holds that the formation of the tumor is due to previous trauma to the area [12]. A medical history of trauma to the site of tumor growth was recorded in one-third of documented cases. According to another theory, because nerve fibers are found in the tumor area with special immunohistochemical techniques, researchers believe that the nerve fibers are stimulated by increased blood flow and release inflammatory prostaglandins. Studies have shown that prostaglandin levels are 100 to 1000 times higher than the levels found in normal bone [13].

In the case we are describing, microscopically, immature osteoid is observed in the larger area, within which few osteocytes are recognized, scattered vascular spaces with a structure like Havers tubes, in the periphery of which few osteoblasts can be distinguished without atypia. Also observed are osteoclasts, scattered chronic lymphoplasmacytic

inflammatory infiltrates, foci of calcification and peripheral changes as reactive fibrosis.

Histochemistry (Masson trichrome, Congo-Red) and immunohistochemistry were performed against: CKAE1-AE3, KP1, PGM1, Vimentin, CD31, CD34, CD44, CD13, CD56 and Ki67 (positive in <1%).

The pulmonary parenchyma adjacent to the tumor shows thickening of the interalveolar septa, the presence of histiocyte macrophage foams within the alveolar spaces, chronic lymphocytic infiltrates around the bronchioles and hyperplasia of type II pneumocytes. Consequently, the morphological and immunophenotypic findings are compatible with a rare osteoid osteoma of the lung.

Surgical excision is the preferred treatment for a pulmonary osteoid osteoma [14,15].

CONCLUSION

Osteoid osteoma is a benign bone tumor originating from osteoblasts and certain components of osteoclasts. Osteoid osteomas are usually less than 1.5 cm in size. The tumor affects any bone in the body, most often long bones such as the femur and tibia. They represent 10 to 12 % of all benign bone tumors. An osteoid osteoma has a good prognosis as it is a benign process without the possibility of malignant transformation. Extraskeletal osteoid osteoma of the lung is an extremely rare medical-pathology entity. Studying the literature, we found that this is only the second case of osteoid osteoma of the pulmonary parenchyma.

REFERENCES

1. Eva Markert , Ulrike Gruber-Moesenbacher, Christian Porubsky, Helmut H Popper: Lung osteoma--a new benign lung lesion. Virchows Arch 2006 Jul;449(1):117-20.

2. Ren X, Yang L, Duan XJ. Three-dimensional printing in the surgical treatment of osteoid osteoma of the calcaneus: A case report. J Int Med Res. 2017 Feb;45(1):372-380.

3. Akinmoladun VI, Akadiri AO, Olusanya AA: Peripheral osteoma of the zygoma: literature review and case presentation. Afr J Med Med Sci. 2007 Dec;36(4):381-383.

4. Cheng J, Garcia R, Smouha E : Mastoid osteoma: A case report and review of the literature. Ear Nose Throat J. 2013 Mar;92(3):E7-9.

5. Chen SM, Chuang CC, TohCH, Jung SM, LuiTN :Solitary intracranial osteoma with attachment to the falx: a case report. World J Surg Oncol. 2013 Sep 8;11:221.

6. Jordan RW, Koç T, Chapman AW, Taylor HP. Osteoid osteoma of the foot and ankle--A systematic review. Foot Ankle Surg. 2015 Dec;21(4):228-34.

7. Gurkan V, Erdogan O. Foot and Ankle Osteoid Osteomas. J Foot Ankle Surg. 2018 Jul-Aug;57(4):826-832.

8. Kato N, Endo Y, Tamura G, MotoyamaT: Multiple pulmonary leiomyomatous hamartoma with secondary ossification.PatholInt. 1999 Mar;49(3):222-5.

9. Lewis VO, Morris CD, Parsons TW. Malignant and benign bone tumors that you are likely to see. Instr Course Lect. 2013;62:535-49.

10. Gryglewski K., Napora J., Wałejko S., Mazurek T. Recent advances on diagnosis and treatment of osteoid osteoma. Chir. Narzadow Ruchu Ortop. Pol. 2022;87:25–31.

11. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg. 2011 Nov;19(11):678-89.

12. Laurence N, Epelman M, Markowitz RI, Jaimes C, Jaramillo D, Chauvin NA. Osteoid osteomas: a pain in the night diagnosis. Pediatr Radiol. 2012 Dec;42(12):1490-1501.

13. Greco F, Tamburrelli F, Laudati A, La Cara A, Di Trapani G. Nerve fibres in osteoid osteoma. Ital J Orthop Traumatol. 1988 Mar;14(1):91-4.

14. Mallepally A.R., Mahajan R., Pacha S., Rustagi T., Marathe N., Chhabra H.S. Spinal osteoid osteoma: Surgical resection and review of literature. Surg. Neurol. Int. 2020;11:308.

15. Bhambhu V., Patel P.G., Mehendiratta D., Dalvie S. Complete Surgical Excision with Pre-operative Localization of Lesion Under CT-Guidance of Osteoid Osteoma of the Sacrum—A Case Report. J. Orthop. Case Rep. 2020;10:56–60..

ΠΑΡΟΥΣΙΑΣΗ ΠΕΡΙΣΤΑΤΙΚΟΥ

Οστεοειδές οστέωμα του πνεύμονα: μια εξαιρετικά σπάνια παθολογική οντότητα

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ΠΕΡΙΛΗΨΗ

Το οστεοειδές οστέωμα περιεγράφη για πρώτη φορά από τον Jaffe το 1935 και αντιπροσωπεύει το 10%-12% όλων των καλοήθων όγκων των οστών. Δεν είναι επιθετικό ούτε έχει τη δυνατότητα κακοήθους εξαλλαγής. Αυτός ο όγκος των οστών συχνά επηρεάζει τα μακρά οστά του μηριαίου οστού και της κνήμης. Το οστεοειδές οστέωμα είναι μικρός, οστεοβλαστικός, καλά οριοθετημένος, καλοήθης όγκος των οστών με τυπικό μέγεθος <1 cm και χαρακτηριστική περιβάλλουσα ζώνη αντιδραστικού οστού. Το εξωσκελετικό οστεοειδές οστέωμα του πνεύμονα είναι εξαιρετικά σπάνιο. Στον πνεύμονα, υπάρχουν όγκοι με οστικό στοιχείο, όπως το αμάρτωμα και ο αμυλοειδής όγκος, και μερικές εξαιρετικά σπάνιες αντιδραστικές αλλοιώσεις όπως η οστική μεταπλασία. Η παρούσα εργασία περιγράφει μια εξαιρετικά σπάνια περίπτωση οστικού οστεώματος του πνεύμονα. Μελετώντας τη βιβλιογραφία, διαπιστώσαμε ότι αυτή είναι μόνο η δεύτερη περίπτωση οστεοειδούς οστεώματος του πνευμονικού παρεγχύματος.

Λέξεις ευρετηρίου: οστεοειδές οστέωμα, πνεύμονας

Ν. Μπαλταγιάννης, Μ. Μπαλταγιάννη, Γ. Στάνκ, Ε. Σούκα, Α. Παγουλάτου, Τ. Παπαδοπούλου, Χ. Βαλαβάνης, Δ. Αναγνωστόπουλος, Ν. Μπολάνος. Οστεοειδές οστέωμα του πνεύμονα: μια εξαιρετικά σπάνια παθολογική οντότητα. Επιστημονικά Χρονικά 2024; 29(2): 271-276

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