Gaziantep Üniversitesi Tıp Fakültesi Dergisi, 4:277-282, 1993

HYPERTROPHIC PULMONARY OSTEOARTROPATHY PIERRE-MARIE-BAMBERGER DISEASE CASE REPORT

Erol ERGÜLER, Ali N SAVAŞER**, Levent ELBEYLİ***, Nüvit ÖZMEN**, Claudia SAVAŞER*****

Key Words:Hypertrophic Osteoartropathy, radioisotope scaning, pulmoner carsinom
Anahtar Terimler:Hipertrofik osteoartropati, radyoizotop sintigrafi, pulmoner karsinom

SUMMARY

Hypertrophic Pulmonary Osteoartropathy (HPO), in particular, is a pulmonary cancer clinical case. An early diagnosis would enable to operable cancer period. This case can be detected through radioactive scanning. A clinical epidermoid carsinom case of a patient is presented in this paper.

OZET

Hipertrofik Pulmoner Osteoartropati, Pierre-Marie Bamberger Hastalığı Vaka Takdimi

Hipertrofik pulmoner osteortropati (HPO) özellikle akciğer kanserinde gözlenen bir klinik antitedir. Erken tanımlanması ve gösterilmesi ile kanser operabl dönemde tespit edilebilir.

Kemiklerde radyofarmasötiğin tipik tutulumunun gösterilmesi mümkün olabilmektedir. Kliniğe müracaat edip epidermoid karsinom tanısı histopatolojik olarak konulan ve ameliyat edilen hastayı takdim ediyoruz.

INTRODUCTION

Hypertrophic pulmonary osteoarthropathy (HPO) is the formation of new osteoit cells in distal part of long bones in human body (radius, ulna, fibula, femora, humeri, metacarpals and mertatarses)(1). This finding may be related to nonmetastatic extrathoracic manifestation of lung carsinoma(2). After that, calsification would follow subperiostal osteoid formation. Then, swelling would effect the adjacent tissues, synovium and adjacent joints in body. This finding is observed, through radiographic techniques assesments, to be symmetric. HPO may precede the diagnosis of tumor by several months and may respond prompty to surgical treatment of a lung cancer(3,4).

CASE REPORT

A 65 years old male (J.R.), was admitted to Berlin Heckeshorn Lungenklinik

* *Gaziantep University Department of Nuclear Medicine, Assistant Proffessor*
** *Krankenhaus Zehlendorf. Berlin-Almanya, Department of Nuclear Medicine, MD*
*** *Gaziantep University Department of Thoracic Surger, Assistant Proffessor*
**** *Private Praxis, Berlin-Almanya, MD*

Department of Pulmonologie II for having pulmonary symptoms and pain in his bones. A direct P-A and lateral chest roentgenogram showed compensatrice emphysema in his right lung, and atelectasis in the left lung (Figure 1,2). Conventional chest tomography showed the central tumor to be on the left side. In the Nuclear Medicine department; 100 MBq Tc 99m MAA was given in supine position by intravenous and taken perfusion images. Perfusion scintigraphy showed a perfect lung on the right hand side, while the left hand side showed the lung in a nearly completely perfusion defect (Figure 3). Computer calculations indicated to a 91.1 % and 9.9 % perfusion in the right and left lungs successively. In the following day, 740 MBq Tc-99m-MDP was given by intravenous. Three hours later, full-body bone-scanning was taken, and examined. An increase in activity of femura and tibia were observed in particular. This increase in activity appeared to be in double-striped in long bones (Figure 4). A tumor was observed in CT on the left lung. This tumor appeared to have attacked the left upper lobe, and due to that, the lower lobe was under compression. A poststenotic pneumonia was observed on the lower lobe. No signs of distant metastasis appeared through CT. Bronchoscopic examination showed that there was an external compression on the left main bronchus. Biopsy was taken and histopathological examination showed a epidermoid carsinom. At the same time, blood samples showed Ca 19-9, teofilin, PSA, NSE to be in the normal ranges. Tuberculosis microskopie appeared to be negative. The cultures of blood and bronchoscopic samples were negative. A pneumonectomy and lymphatic disection were done at the same operating sesion. One month later follow up examinations showed the patient to have no complications or significant manifest symptom.

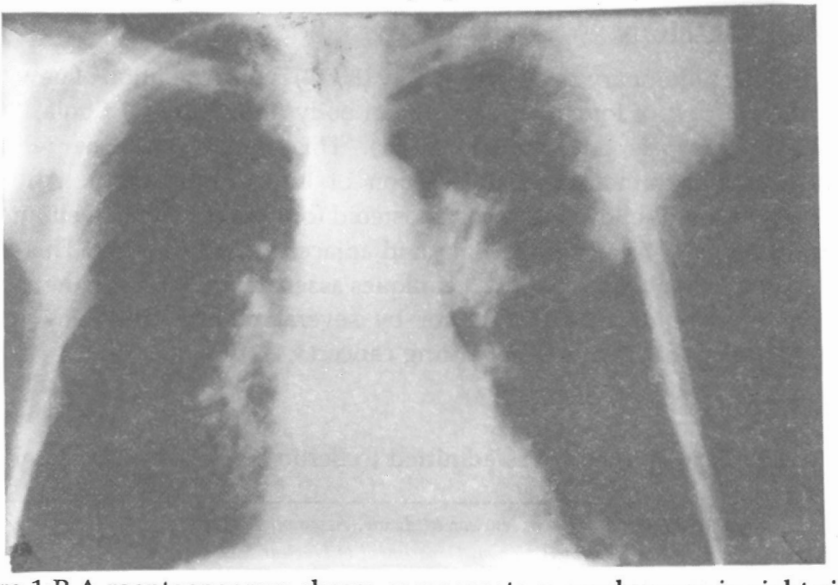

Figure 1:P-A roentgenogram shows compensatory emphysema in right side.

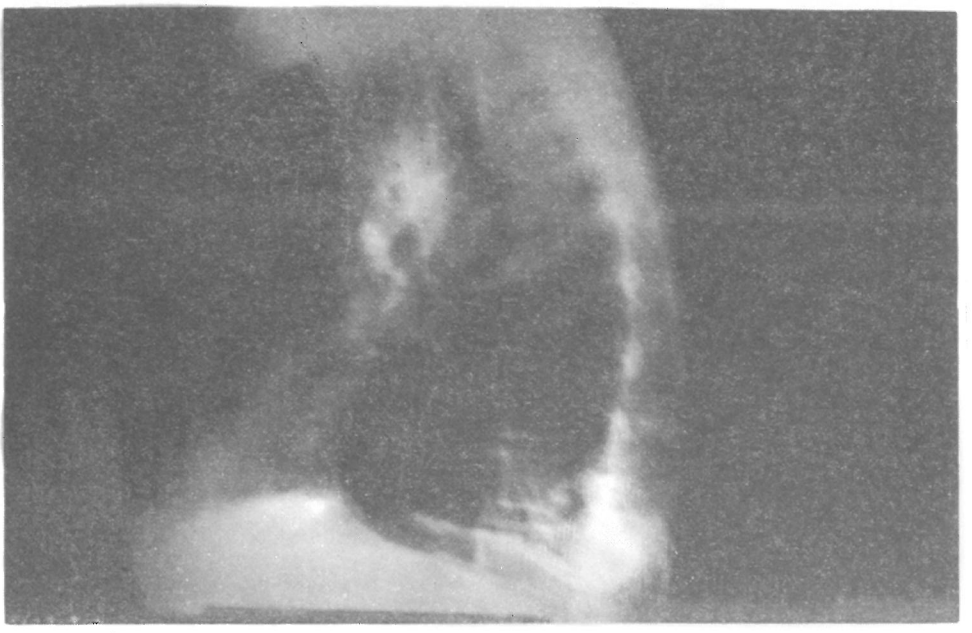

Figure 2:Lateral chest roentgenogram of the same patient.

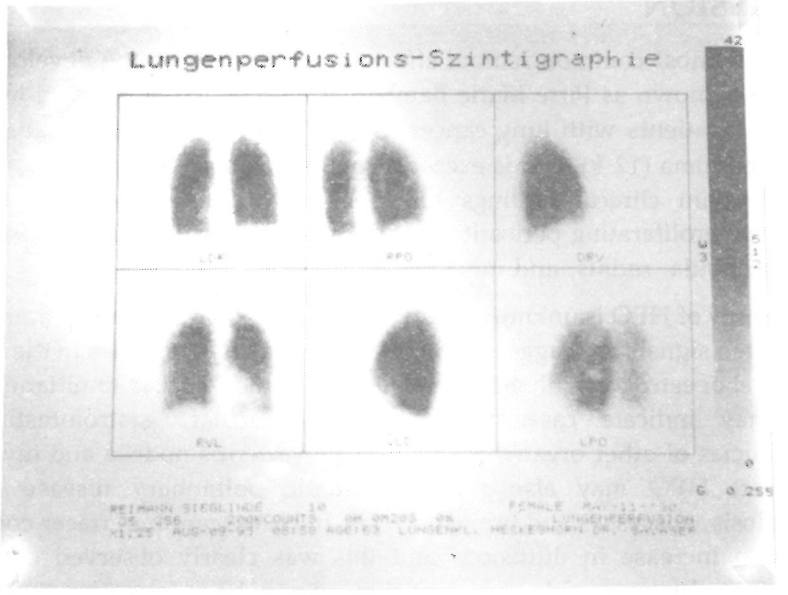

Figure 3:Pulmonary perfusion scintigraphy showed a perfusion defect on the left side.(Preoperative period).

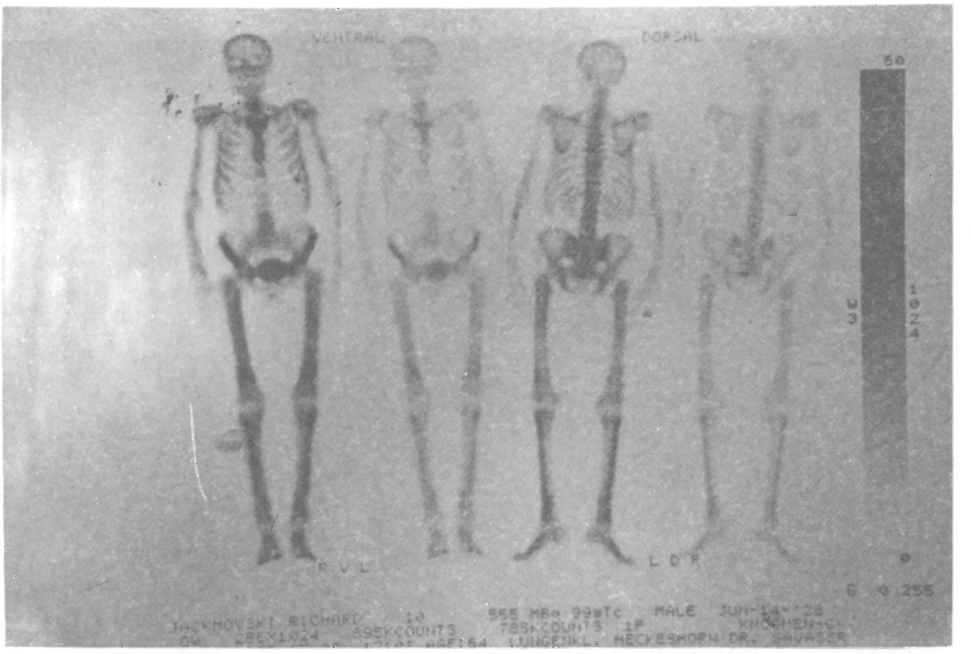

Figure 4:Whole-body bone scintigraphy of a 65 year old man who was admitted because of pulmonary symptoms and pain his bones.

DISCUSSION

One of the most common remote effects of lung cancer is the development of HPO, also known as Pirre Marie Bamberger disease. HPO is found in up to 5-9.2 % of patients with lung cancer, is particularly common in patients with adenocarcinoma (12 %), and is exceedingly rare in small cell lung cancer (3,5,6). The dominant clinical findings are clubbing of the fingers and toes and symmetric proliferating periostitis of the distal ends of long bones, particilarly the tibia, fibula, radius, and ulna.

The etiology of HPO is unknown, and estrogens, growth hormone, and neurally transmitted signals are suggested inconclusively as playing roles in the etiology. High level of estrogen with gynecomastia, are found to occur simultaneously(3). HPO may indicate cases such as cardiovascular, gastrointestinal, the malignancies of other organs, pregnancy, polyarteritis nodosa and myxedema. Moreover, HPO may also indicate chronic pulmonary disease such as tuberculosis, abscesses and bronchiectasis. In bone scanning, tracer counting is known to increase in diffusion, and this was clearly observed during this experiment. The inner side of periost showed new bone formation and periostic thickening. This may also occur simultaneously with a common increase in radius, ulna, the lower part of tibia and the lower part of femur. A comparison

between x-rays and scintigraphy would show HPO(7). Bone roentgenograms show an ossifying periostitis, and the bone scan to be positive in affected bones(3). The increase of circumferancial cortial counting vould be detected, in general, in HPO, paget's disease, and stress fractur(8,9).

Long bones show significant increase in the activity of their cortical margine. The other side of the bone when affected is generally observed to have "double stripe" sign. In most cases, only long bones are known to be affected in their juxta-articular part, especially falanx(10). In such cases, metastatic disease may be mistakenly diagnosed instead of systemic bone lesion and vice versa. Scapula, mandibula, maxilla, clavicula and patella are, generally effected. HPO may detect the bilateral effect on the patella, which indicates to an increase in chondromalasia and degenerative arthritis(11). The bone changes of hyperparathroidism can simulate HPO. A bone roentgenogram may show normal signs while radioisotope scanning tests are pathological previous. The subperiostal new bone formation may be related to previous hyperemia(12). Such condition causes severe pain. The artropathy may be extremely disabling and painful, and treatment is often ineffective(4,13).

The signs observed in the carsinoma of the lungs would disappear after resected the tumor(3,14). Hypertrophic pulmonary osteoarthropathy may be the only way to show symptoms of lung cancer at an early stage. Therefore, the information obtained through HPO may be considered valuable. The authors believe that radionuclide bone scanning is a successful techniques in HPO detection.

REFERENCES

1- Smith LH.:Systemic manifestations of carsinoma of the Lung. In Murray JF., Nadel JA.:Textbook of Respiratory Medicine. WB Sounders Company 1988 pp 414-430.

2- Meade P., Moad J., Fellows D., Adams CW.:Carsinosarcoma of the lung with hypertrophic pulmonary osteoarthropathy. Ann.Thorac.Surg.1991;51:488-90.

3- Roth JA., Ruckdeschel JC., Weisenburg TH.:Thoracic Oncology Section 2.Diagnosis and Staging. Chapter 5.Clinical Presentation of Lung Cancer. LA Doyle., J Aıser WB Sounders, Philadelphia. 1989, pp 70-71.

4- Blackwell N., Bangham L., Hughes M., Melzack D., Trotman I.:Treatment of resistant pain in hypertrophic pulmonary artropathy with ketorolac. Thorax 48:401, 1993.

5- Grech P., Marten TJ., Barrington NA., et al.:Diagnosis of metabolic bone disease. Chapter 10. Radionuclid studies in metabolic disease of bone. Philadelphia, Sounders, 1985 pp 298-371.

6- Stenseth JH., Clegett OT., Woolner LB.:Hypertrophic pulmonary osteoartropathy. Dis Chest 52:62-68, 1967.

7- Fogelman and Maisey. An Atlas of Clin.N.M.Deutscher Arzte-Verlag Kõln 1988 pp 64-66

8- Citrin DL., Mc Killop JH.:Atlas of Tc bone scans. Philadelphia:W.B.Sounders 1978.

9- Spencer RP.,Levinson ED., Baldwin RD.,et al.:Diverse bone scan abnormalities in "shin splints". J Nuc Med 20:1271-1272, 1979.

10- Ali A., Tetelman MR., Fordham EW., et al.:Distribution of hypertrophic pulmonary osteoarthropathy. AJR 134:771-780, 1980.

11- Kipper MS., Alazroki NP., Feiglin DH.:The "hot" patella. Clin Nucl Med 7:28-32, 1982.

12- Costello P., Gramm HF., Lokich J.:Detection of hypertrophic pulmonary osteoarthropathy associated with pulmonary metastatic disease. Clin Nucl Med 2:397-99, 1977.

13- Freeman MH., Tonkin AK.:Manifestations of hypertrophic pulmonary osteoarthropathy in patients with carsinoma of the lung. Radiology 120:363-365, 1976.

14- Carrol KB., Doyle L.:A common factor in hypertrophic osteoarthropathy.Thorax 29:262-264, 1979.