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Case Report

# Bone Scintigraphy for Diagnosing Atypical Chest Pain of Musculoskeletal Origin

**Soo-Young Kim and Jeong-Gil Leem***

Department of Anesthesiology and pain Medicine, University of Ulsan College of Medicine, Republic of Korea

***Corresponding author:** Jeong-Gil Leem, Department of Anesthesiology and pain Medicine, Asan medical Center, University of Ulsan College of Medicine 88 Olympic-ro 43-gil, Songpa-gu, Seoul, 05505, Republic of Korea

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## Abstract

Bone scintigraphy is a commonly used imaging technique for assessing abnormal bone metabolism. In this article, we review 3 cases of patients with atypical chest pain of musculoskeletal origin. First, in the case of Tietze's syndrome, bone scintigraphy showed the focal uptake of costochondral junction of the first rib consistent with clinical symptoms and played a key role in diagnosis. Second, in the case of delayed union sternum fracture, bone scintigraphy did not only help in diagnosis, but it also suggested that clinicians can assess the state of bone healing with time. Finally, in the case of suspected Synovitis-Acne-Pustulosis-Hyperostosis-Osteitis (SAPHO) syndrome, bone scintigraphy was useful for evaluating the cause of pain. From these cases, we concluded that bone scintigraphy is a practical tool for evaluating musculoskeletal cases with atypical chest pain and monitoring the change in bone metabolism as diseases progress or resolve.

**Keywords:** Acquired hyperostosis syndrome; Bone; Chest pain; Fracture; Musculoskeletal diseases; Pain clinics; Radionuclide imaging; Tietze's syndrome

**Abbreviations:** SAPHO: Synovitis-Acne-Pustulosis-Hyperostosis-Osteitis; CRPS: Complex Regional Pain Syndrome; CT: Computed Tomography; MRI: Magnetic Resonance Imaging; NRS: Numeric Rating Scale; ESWT: Extracorporeal Shockwave Therapy

## Introduction

Bone scintigraphy is a widely used nuclear imaging technique for detecting osteoblastic activity [1,2]. A radioisotope such as Technetium-99m is absorbed by hydroxyapatite crystals on mineralizing bone surfaces and revealed as "hot spots" or "cold spots", showing abnormal bone metabolism. "Hot spots" indicate excessive radioactive substance, while "cold spots", on the other hand, indicate a lack of blood supply. Using bone scintigraphy, clinicians can detect bone infection or inflammation, which are not detected on a simple x-ray; hence, it is commonly used for diagnosing various bone disorders [3]. Oncologists can also detect metastatic lesions of patients with malignancies because one of the strengths of bone scintigraphy is visualizing the whole body [2]. Additionally, bone scintigraphy is important in the pain clinic. First, bone scintigraphy helps to correlate clinical symptoms with bone lesions in patients with low back pain. Low back pain, which is a very common complaint in the pain clinic, has various causes such as benign or malignant bone tumors, osteoporotic vertebral compression fractures, facet joint arthropathy, and spondylolysis. Computed Tomography (CT) or Magnetic Resonance Imaging (MRI) also shows morphologic changes in the bones and the joints, but these findings are also shown in asymptomatic patients. Thus, findings of bone scintigraphy are sometimes more specific than those of CT or MRI in evaluating low back pain. Second, when pain physicians suspect Complex Regional Pain Syndrome (CRPS), bone scintigraphy may provide clues for diagnosis. CRPS is a very complicated disease that is difficult to diagnose with only one tool. Although bone scintigraphy findings are not included in the revised diagnostic criteria of CRPS by the International Association for the Study of Pain, it provides additional information for assessing patients. Finally, patients who have pain in the extremities usually have musculoskeletal or rheumatoid diseases. In these patients, as stated above, clinicians can observe the abnormal bone metabolism related to pain by using bone scintigraphy [4]. In the present case series, we report 3 cases that used bone scintigraphy to guide diagnosis and treatment in patients with atypical chest pain; this provides insights into its effectiveness as a diagnostic tool for uncommon cases.

### Case 1: Tietze's Syndrome

A 53-year-old woman presented for evaluation of a right subclavicular pain. The pain began 3 years ago without any trauma history. Furthermore, it had been aggravated 1 month earlier, and

the pain radiated to her right scapular area. She could not rotate her shoulder because of the pain. The intensity of pain was 3 or 4 points on the Numeric Rating Scale (NRS), and it was getting worse. She also had a common cold, but she found it difficult to cough because of the pain. In addition, there was no relieving factor. Physical examination revealed mild swelling and tenderness around her right clavicle. She was suspected of having Tietze's syndrome without any radiographic imaging at another hospital 3 years earlier, and she took medication for pain control. She also took steroids intermittently, when the pain increased, by doctor's prescription. Despite several treatments, the pain increased 1 month ago and radiated to her right back. We recommended bone scintigraphy to rule out other possible reasons for the aggravating pain.

The bone scintigraphy in our hospital showed a focal hot uptake at the right costochondral junction of the first rib, which suggested Tietze's syndrome. Chest CT also showed mixed lytic, sclerotic lesions and enlargement in the right 1st rib. Follow-up CT was indicated for ruling out sclerosis associated with costochondritis or Tietze's syndrome (Figure 1). We concluded that Tietze's syndrome was the most probable diagnosis based on bone scintigraphy findings. Oral medications such as celecoxib and methotrexate that had been prescribed by her rheumatologist did not relieve the pain enough, and we decided to perform an Extracorporeal Shockwave Therapy (ESWT) when she first visited our clinic. It was effective, and it was repeated 3 times every week. Her pain was relieved, and she was well for approximately 1 month. However, her pain aggravated again, and she visited our clinic 1 month after the last ESWT. At that time, she underwent these therapies twice a week, but it did not work as the previous sessions and she wanted another treatment. We planned intercostal nerve block with lidocaine and dexamethasone. After this procedure, the pain decreased.

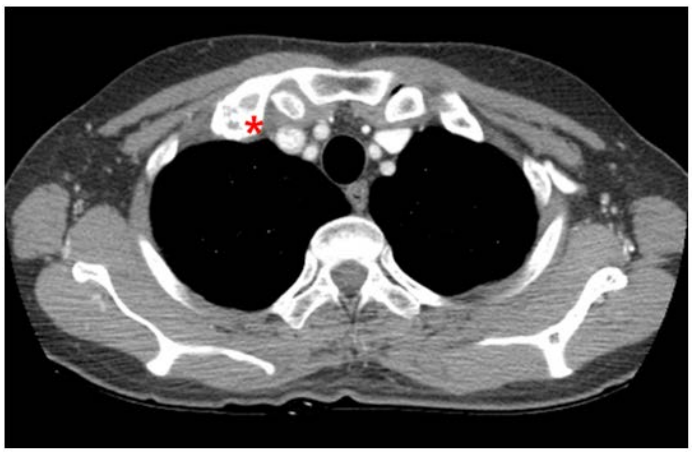

**Figure 1:** Chest CT shows mixed lytic, sclerotic lesions and enlargement in the right 1st rib to rule out sclerosis associated with costochondritis or Tietze's syndrome (Asterisk).

**Case 2: Delayed Union Sternum Fracture**

A 63-year-old man with left anterior chest pain reported to our clinic. The pain began after a traffic accident that happened 8 months earlier, and he was diagnosed with a depressed sternum fracture at that time. He was assured that the fracture would heal spontaneously, and the pain would also resolve. However, his pain persisted for a month after the accident, and he underwent chest CT and echocardiography to rule out cardiac and pulmonary disorders; no abnormalities of the lungs and the heart were found. He took oral medications, but it did not work. Although 8 months had elapsed after his accident, the pain persisted and that was the reason for reporting to our clinic. The intensity of pain was between 3 and 8 points on NRS. It was aggravated when he worked at the office, and there was no relieving factor. On physical exam, he had tenderness at the sternum. Results of laboratory tests, including complete blood count test, C-reactive protein, and erythrocyte sedimentation rate, were normal.

Bone scintigraphy revealed horizontal increased uptakes in the sternum body, which was indicative of a nonunion sternum fracture. We also found a fracture line in his sternum on ultrasound (Figure 2A). He underwent ESWT on his first visit and a distal intercostal nerve block with 0.5% bupivacaine on his second visit, which was 2 weeks after the first visit. Both were effective. However, he returned after 2 months due to recurrent pain and was administered with a subperiosteal injection of 0.25% bupivacaine at the fracture site under ultrasonography. He also underwent ESWT one more time. Two months after his re-visit, he reported to our clinic for the fourth time because of aggravating pain. We performed a distal intercostal nerve block with 0.5% bupivacaine and suggested alcohol neurolysis if it worked enough. This meant that he expected a pain relief of more than 70%. However, on the next visit, he reported that he observed only 50% relief initially, which returned to 0% after 2 or 3 days.

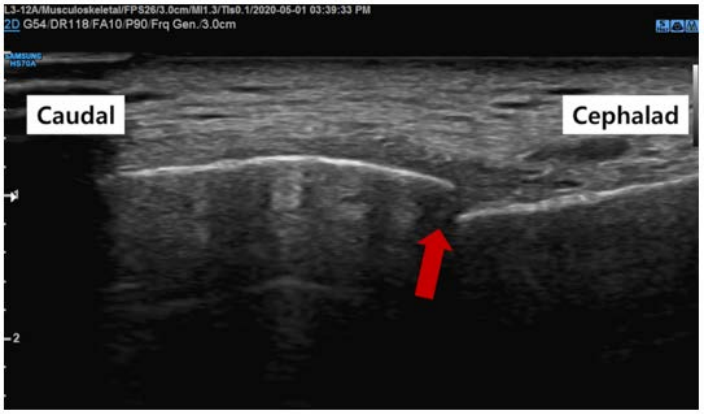


**Figure 2A:** Fracture line in sternum on ultrasound (Red arrow).

Therefore, we consulted a thoracic surgeon to consider

surgical fixation of the sternum fracture, and he recommended a chest CT. The CT scan showed a fracture at the body of the sternum and scanty callus formation (Figure 2B). Additionally, a radiologist observed healing of the sternum fracture with deformity, and bone scintigraphy at follow-up was indicated. The thoracic surgeon opted to observe him and request follow-up CT and bone scintigraphy after 6 months. We also consulted an endocrinologist for the appropriateness of Teriparatide therapy, which is a form of parathyroid hormone that accelerates bone union. It is currently being considered to speed up bone healing.

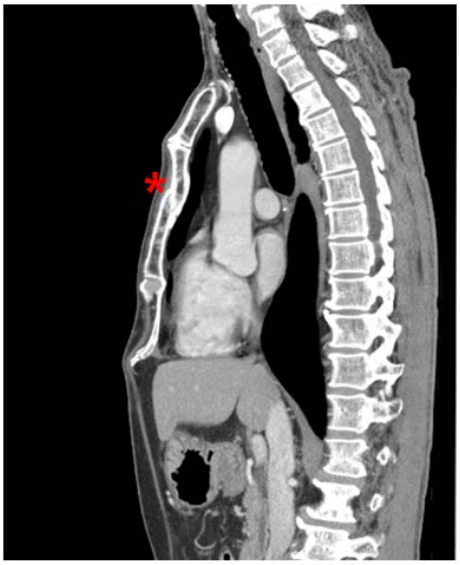

**Figure 2B:** Chest CT shows a fracture in the body of the sternum and scanty callus formation (Asterisk).

**Case 3: SAPHO Syndrome**

A 65-year-old woman was referred from another hospital for right chest pain around her right nipple. The pain had begun spontaneously 3 years earlier. She visited a local orthopedic clinic and met a cardiologist and thoracic surgeon, but none of them could ascertain the cause of the pain. The pain was aggravated on leaning, coughing, and lying down, and it was relieved when she spread out her chest. The intensity of pain was 3 points on NRS, and physical exam revealed tenderness around her nipple. We requested bone scintigraphy and chest CT. The result of bone scintigraphy showed arthritis in the sternomanubrial and right sternoclavicular joints, and SAPHO syndrome had to be ruled out (Figure 3A). Chest CT showed degenerative changes with sclerosis and subtle subchondral cyst formation at the sternum, the manubriosternal joint, and the right sternoclavicular joint, which suggested osteoarthritis (Figure 3B). There was no remarkable hyperostosis at the sternoclavicular joint, which was a clinical feature of SAPHO syndrome. It is also known as acquired hyperostosis syndrome. However, there was an opinion on the sternoclavicular joint view of the chest CT that was consistent with SAPHO syndrome.

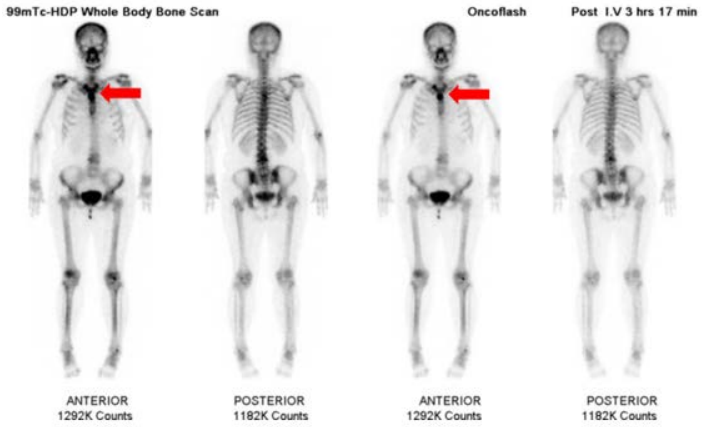


**Figure 3A:** Bone scintigraphy shows arthritis in the sternomanubrial and right sternoclavicular joints (Red arrow).

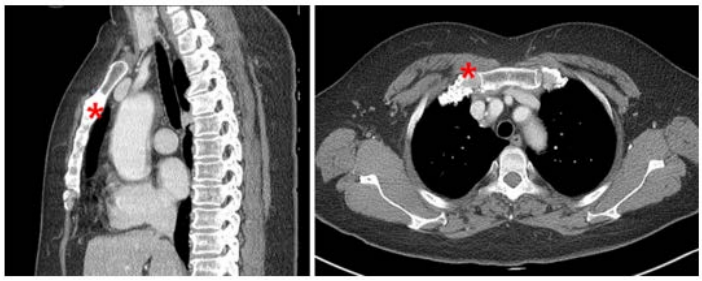

**Figure 3B:** Chest CT shows degenerative changes with sclerosis and subtle subchondral cyst formation at the sternum, manubriosternal joint and right sternoclavicular joint (Asterisk).

We thought that SAPHO syndrome was more probable than costochondritis based on these findings, and we needed more evidence for confirmation. Thus, while managing her pain, we consulted with other departments. For pain control, she underwent ESWT twice in 2 weeks and took oral medication. The intensity of the pain reduced to 1 point on NRS. However, her pain aggravated without any specific event after 3 months; it originated from her left chest. She reported that the pain alternated between both sides and sometimes it moved to her back. We administered a sternomanubrial injection of 0.5% bupivacaine and triamcinolone, and it worked for a few days. We also consulted a gastroenterologist to rule out reflux esophagitis, a rheumatologist to rule out arthritis, and a thoracic surgeon and a cardiologist to rule out other cardiopulmonary diseases. In conclusion, she was suspected of reflux esophagitis and sternomanubrial and sternoclavicular joint arthritis. Thus, she planned to visit them periodically. We are going to observe her and she has been scheduled for regular follow-ups. As her symptoms change, we will perform further imaging studies or laboratory tests to confirm her diagnosis.

## Discussion

We have demonstrated that bone scintigraphy may be useful for diagnosing and ascertaining the cause of aggravating pain when clinicians encounter patients presenting with atypical chest pain.

First, bone scintigraphy provides clues for diagnosing atypical diseases that present with chest pain such as Tietze's or SAPHO syndromes. Common causes of chest pain are ischemic heart disease, Herpes zoster, and gastroesophageal reflux disease [5,6]. Patients with ischemic heart disease can be easily diagnosed by echocardiography, electrocardiogram, or serum cardiac markers such as troponin-I and creatinine kinase. Patients with specific unilateral lesions and medical history, who report frequently in the pain clinic, can be easily diagnosed with Herpes zoster. However, unusual cases such as Tietze's syndrome or SAPHO syndrome often lack definite proofs, and bone scintigraphy may be considered because it can detect abnormalities of bone metabolism in the area patients feel pain. The positive result of bone scintigraphy suggests that a patient has a musculoskeletal problem [1]. Furthermore, bone scintigraphy is more readily available and cheaper than CT or MRI for initial screening of these atypical diseases.

Second, we can observe the course of the disease using bone scintigraphy. It provides insights on the state of bone metabolism 1, and enables us to find the reason for aggravating pain. We can also judge the efficacy of treatment using bone scintigraphy by comparing findings after treatment to initial findings. Clinicians can judge the effect of chemotherapy or radiotherapy in cancer patients with bone metastasis, osteomyelitis, or arthritis [7,8]. In the second case of sternum fracture, the patient got better for the first few months but got worse again. Chest CT showed healing of the sternum, and follow-up bone scintigraphy was recommended to check the status of the lesion. If the management of pain is appropriate, follow-up bone scintigraphy will not show the abnormality in the lesion. In this regard, when we want to know the effect of treatment or consider changing the treatment, bone scintigraphy may be helpful for decision making.

Third, clinicians can detect a hidden malignancy easily with bone scintigraphy. As we stated above, one of the strengths of bone scintigraphy is the visualization of the whole body. Patients with chest pain may not only have local lesions within the chest; they may also have metastatic lesions in other areas due to unknown malignancy [5,9]. This emphasizes the importance of ruling out malignancies when we first encounter patients with atypical chest pain. Although we are not able to diagnose cancer with bone scintigraphy alone, we can find out if hidden malignancies exist or not.

Despite the advantages described above, the specificity of bone scintigraphy is not that high although the sensitivity is high. The sensitivity of bone scintigraphy is almost 100% for detecting stress fractures; however, it shows the rate of new bone formation in general and subtle early changes in bone metabolism, making its specificity low [10-13]. This is why it is difficult to diagnose certain diseases only with bone scintigraphy. In addition, bone scintigraphy uses radioisotopes, and clinicians should consider the time it takes and the risk of radiation exposure. Thus, clinicians should carefully consider the risks and benefits of bone scintigraphy when they manage patients presenting with atypical chest pain of suspected musculoskeletal origin.

Bone scintigraphy may be helpful for the evaluation of musculoskeletal conditions with atypical chest pain. It may also help clinicians to ascertain the causes of aggravating pain or recognize the course of the disease and reveal hidden malignancies.

## Conflict of Interest

I hereby disclose all of my conflicts of interest and other potentially conflicting interests, including specific financial interests and relationships.

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