

Scintigraphic history of arthritis in whipple's disease

Abstract

The article describes a 4year clinical observation of a 56year-old male with Whipple's disease. The diagnosis was established based on the progressive diarrhea with malabsorption, detection by CT a low-density retroperitoneal infiltrate and mesenteric lymphadenopathy, abundant PAS-positive macrophages in the biopsy specimens of the small intestine and mesenteric lymph nodes. Arthropathy had started 10years before the appearance of intestinal symptoms, regressed with its development and resumed when clinical remission was achieved.

Keywords: whipple's disease, arthritis, retroperitoneal lymphadenopathy, osteoscintigraphy, tc-99m, sodium pyrophosphate, accumulation coefficient

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Introduction

We performed osteoscintigraphy with quantitative assessment of radionuclide accumulation in joints 3 times in periods of different disease activity. Results demonstrated a good correlation with clinical data. In this case scintigraphy also confirmed the efficacy of treatment.

Whipple's disease is a relatively rare disease (since 1907, no more than 1,000 cases have been described). It is a chronic generalized infection caused by the bacterium *Tropheryma whippelii*, close to actinomycetes. Mortality in this disease is still high (up to 25%, despite treatment). Articular involvement occurs in 80% of patients. The most characteristic clinical variant is palindromic rheumatism.^{1,2} There is often an inverse correlation between intestinal and articular symptoms in this disease.³

Patient G, 56years old was admitted for the first time with complaints of a pronounced general weakness, fatigue, decreased appetite, liquid stool up to 3times a day, a periodic body temperature elevation up to 37.7°C, a decrease in body weight by 13kg during the previous year.

Since 2000 he was suffering from the short-term morning episodes of pains in the cervical spine, which resolved without treatment. In 2003, the patient had attacks of pains and swelling in the small joints of the hands and feet, knee and ankle joints. The intensity of such events increased, the pain was so severe that the patient struggled to move around the apartment, and was unable to work. During the interictal period, the symptoms regressed, but the swelling of the metacarpophalangeal joints of both hands persisted. Since May 2004 diarrhea worsened, but arthritis decreased. In December 2004 polyarthritis completely regressed, but weakness increased and work capacity decreased. Since the beginning of 2005, the patient noticed morning stiffness, recurring pains in the small joints of hands and feet, in wrists, in the right claviculo-acromial, knee and ankle joints. The symptoms were lasting 2-3days and spontaneously resolved.

On admission: skin was clean, pale, with a grayish tinge. Heart rate 80beats/min, BP 120/80mm Hg. There were hypochromic anemia (hemoglobin 104g/l, color index-0.73), moderate thrombocytosis ($465 \times 10^9/l$), and elevation of ESR (43mm/h) in laboratory tests. The coprology examination revealed digested and undigested muscle fibers. CT scan of the abdominal cavity demonstrated an extended retroperitoneal mass with polycyclic contours, located behind and

down from the pancreas and near mesenteric vessels and the root of the mesentery; marked enlargements of retrocrural and retroperitoneal lymph nodes. This mass and lymph nodes were of unusual low density.

In the biopsy samples of this mass inflammatory infiltration was determined, represented mainly by macrophages with a PAS-positive foamy cytoplasm. In a biopsy specimen from the duodenum also accumulations of PAS-positive macrophages were found in significantly enlarged villi.

In view of the clinical, radiological and histological signs, Whipple's disease was suspected. The patient was treated with ciprofloxacin 1g/day orally. From the 2nd day of treatment, the stool decreased to 1-2times a day, the consistency of the stool was normalized, weakness decreased, and by the end of the week the body weight gained 3kg. In the next 6months he took trimethoprim/sulfamethoxazole (960mg/day 2weeks) and erythromycin (1g/day 1week) in turn.

In December 2005 the patient felt himself well. He had no complaints, gained 20kg in weight. All laboratory tests were normal as well as the abdominal CT. But in February 2006 articular pain resumed. Osteoscintigraphy with TC-99m pyrophosphate sodium was performed 03.03.2006 (Figure 1).

There was increased accumulation of radiotracer in the projection of shoulder joints, wrist and ankle joints, small joints of hands and feet. In May 2006, the patient stopped taking antibiotics. For about one year he felt himself satisfactory. Gradually, however, the symptoms grew. In November 2007 the patient complained of unformed stool 1-2times a day, bloating, rumbling in abdomen; pain in wrist joints with swelling of periarticular tissue, pain in ankle joints, small joints of the hands and feet without swelling, pain in the cervical and lumbar spine.

Laboratory tests showed only a mild leukocytosis $10.1 \times 10^6/l$, and the increased proportion of neutrophils (88.0%). Biopsy of the duodenal mucosa revealed the moderate atrophy of villi. In submucosal layer the separate groups of PAS-positive macrophages were again detected. Performed computed tomography of the abdomen showed no changes compared with the study from 12.12.2005. In the control of planar osteoscintigraphy (27.11.2007) (Figure 2), however, there were a positive dynamics. In comparison with the previous study there was no increased accumulation of the radiopharmaceutical in the left ankle. Accumulation in the right ankle, the left wrist joint, small joints of the hands, thoracic spine and both sacroiliac joints markedly

decreased. At the same time moderately increased accumulation of the radiopharmaceutical in the right wrist joint. We calculated accumulation index (AI) as a ratio of the activity over the joint and the activity over the adjacent to the joint identical zone over bone. Data are presented in Table 1.

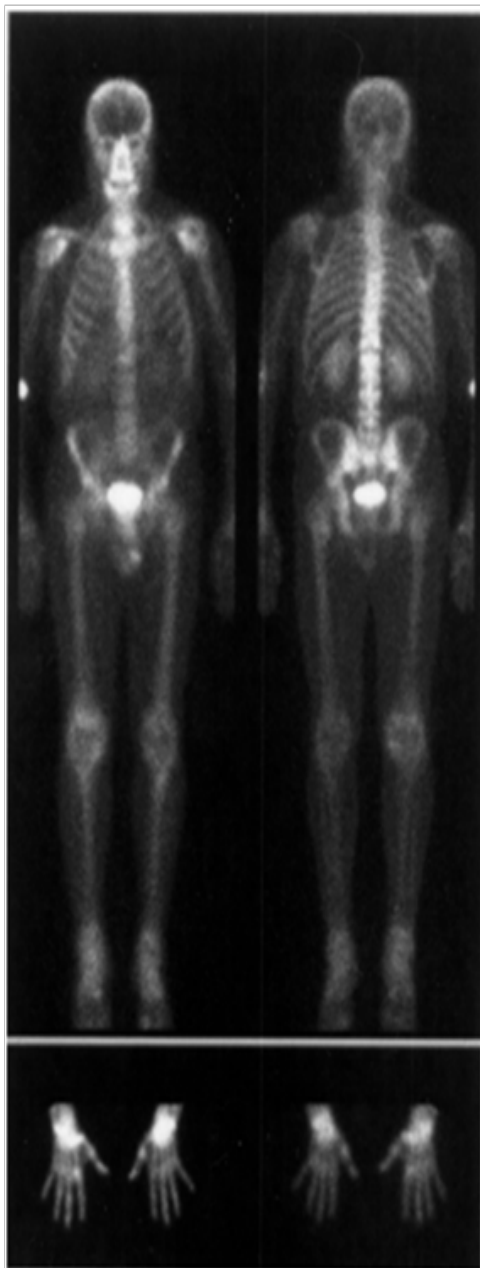


Figure 1 Osteoscintigraphy with TC-99m labeled sodium pyrophosphate (03.03.2006).

The patient was thoroughly followed up. In 2008, the patient was examined in a state of complete clinical remission. Osteoscintigraphy revealed almost complete normalization of the radiotracer accumulation in the joints (Figure 3) (Table 1). The AI of the right elbow decreased by 30% compared to the previous study, AI of shoulder joints decreased by 30%, AI of the right ankle by 43%, tarsal joints-47%.

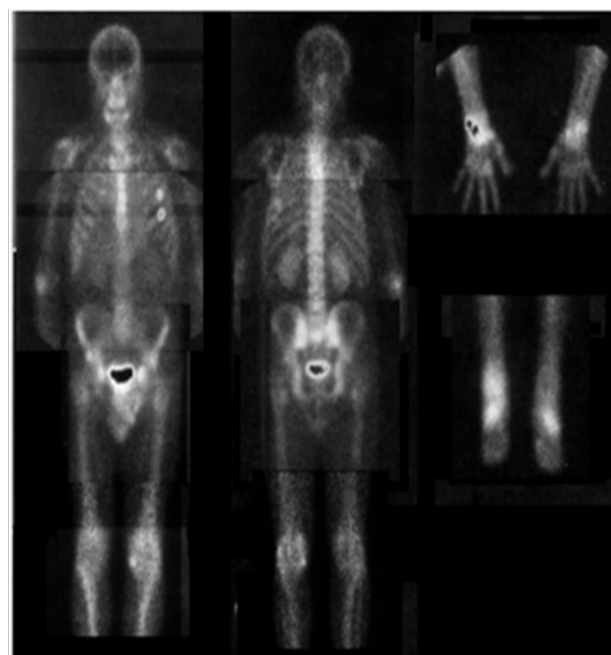


Figure 2 Planar osteoscintigraphy with TC-99m labeled sodium pyrophosphate (27-11-2007).

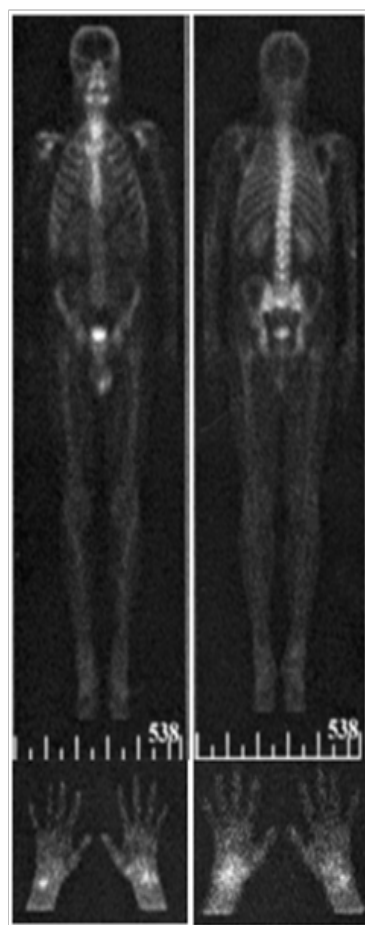


Figure 3 Osteoscintigraphy with TC-99m labeled sodium pyrophosphate (16.09.2008).

Table 1 Values of accumulation index in joints and articulations at 27.11.2007 and at 2008

No	Joint or articulation	Reference values	27.11.2007 (Left/Right)	16.09.2008 (Left/Right)
1	Elbow	1,51	-/2,36	1,74/1,74
2	Knee	1,59	1,58/1,35	1,36/1,33
3	Hip	1,7	1,42/1,46	1,54/1,43
4	Wrist	1,24	1,7/2,29	1,26/1,34
5	Shoulder	1,74	2,16/2,11	1,62/1,79
6	Ankle	1,3	1,54/2,49	1,27/1,42
7	Tarsal	1,2	2,37/2,68	1,50/1,42
12	Sacroiliac	1,24	1,00/1,06	1,0/0,97
13	Sternoclavicular	1,27	1,89/1,9	1,35/1,69
14	Claviculoacromial	1,66	0,75/1,12	0,7/1,03

Abnormal values marked with bold font

Conclusion

In the presented case we observed opposite dynamics of intestinal and articular manifestations of Whipple's disease that was confirmed not only by clinical, laboratory data, but by the results of quantitative scintigraphy. Osteoscintigraphy with the calculation of AI in the joints can be an important quantitative method for objective assessment disease activity of the joints and it may be used to confirm the effectiveness of the treatment in Whipple's disease.

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None.

Conflict of interest

Author declares that there is no conflict of interest.

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