Scintigraphic history of arthritis in Whipple’s disease

Abstract

The article describes a 4 year clinical observation of a 56 year-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 mesenterial lymphadenopathy, abundant PAS-positive macrophages in the biopsy specimens of the small intestine and mesenterial lymph nodes. Arthropathy had started 10 years 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

Introduction

We performed osteoscintigraphy with quantitative assessment of radionuclide accumulation in joints 3 times in periods if 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. There is often an inverse correlation between intestinal and articular symptoms in this disease.

Patient G, 56 years old was admitted for the first time with complaints of a pronounced general weakness, fatigue, decreased appetite, liquid stool up to 3 times a day, a periodic body temperature elevation up to 37.7°C, a decrease in body weight by 13 kg 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 intertical 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 the metacarpophalangeal joints of both hands persisted. Since May 2004 diarrhea worsened, but arthritis decreased. In December 2004 the metacarpophalangeal joints of both hands persisted.

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

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Laboratory tests showed only a mild leukocytosis 10.1 × 10⁹/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.

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%.
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               | 1.00/0.97               |
| 13 | Sternoclavicular      | 1.27             | 1.89/1.9                | 1.35/1.69               |
| 14 | Claviculoacromial     | 1.66             | 0.75/1.12               | 0.71/1.03               |

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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Conflict of interest

Author declares that there is no conflict of interest.

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