

A Dental Clinical Study for 12 Cases of SAPHO Syndrome with Biomedical Research on Trace Elements

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Abstract: SAPHO syndrome is characterized by bone-inflammation with or without skin lesions. We reported about clear effect of oral-treatments for skin lesion of palmoplantar pustulosis. This time, for a first investigation into a relation between oral diseases and pathogenesis of SAPHO syndrome, we made a clinical study for oral-treatments on 12 patients of SAPHO syndrome with palmoplantar pustulosis and biomedical research on trace elements in blood. Dental treatments(treatment for dental chronic focuses, dental metal removal by patch test) were done for 12 patients of SAPHO syndrome with skin, bone and joint symptoms. By the dental treatments, the skin lesion had been cured or decreased remarkably in all cases, and pain of bone and joints had been cured in all cases. The blood samples from 7 patients with SAPHO Syndrome were analyzed for trace metals. The analyzed trace metals were serum zinc serum copper serum iron blood selenium and blood mercury. The serum copper level was significantly high, the serum zinc level was significantly low and the blood selenium level was significant low than healthy control. The other trace metals in the blood showed no remarkable change. These result suggests a possibility intra-oral-focus, dental-metals and trace metals has some relation to pathogenesis of SAPHO syndrome.

Keywords: SAPHO syndrome, palmoplantar pustulosis, dental treatment, trace metals

Introduction

Synovitis, Acne, Pustulosis, Hyperostosis, Osteitis syndrome (SAPHO syndrome) is advocated by Chamot and Kahn et al.¹⁾ in 1987 as a general disease by unknown-pathogenesis. This syndrome is characterized by bone-inflammation with or without skin lesions, palmoplantar pustulosis, pustular psoriasis and severe acne (Table 1).

We reported about the clear effect by oral-treatments for improvement of skin symptoms on palmoplantar pustulosis²⁾. otherwise, recently the relation between trace metals in blood samples and some difficult treatment skin diseases such as psoriasis and pemphigoid include autoimmune diseases was suggested. This time, for a first investigation into a relation between oral diseases and pathogenesis of SAPHO syndrome, we made a

clinical study for oral-treatments on 12 patients of SAPHO syndrome with palmoplantar pustulosis and biomedical research on trace elements.

Materials and Methods

Patients:

12 patients of SAPHO syndrome with palmoplantar pustulosis who visited, treated and observed in our dental hospital our department from Apr. 1987 to July 2003 were studied. They were diagnosed by dermatologist and orthopedist, all cases were positive by ^{99m}Tc bone scintigraphy. The age range was from 29yr. old to 56yr. old, an average of 42.3yr. The ratio of men to women was 1:3.

The blood samples from 7 patients with SAPHO Syndrome were analyzed for trace metals. The age range was from 18yr. old to 72yr. old, an average of 42.8yr. old. The ratio of men to women was 4:1. The blood samples from 15 healthy persons, staffs in our university, were analyzed for healthy control. The age range was from 27yr. old to 62yr. old, an average of 38.29yr. old±10.95yr.old. The ratio of men to women was 7:8.

Chronic recurrent multifocal osteomyelitis Usually sterile* Spine may be involved With or without skin lesions
Acute, subacute, or chronic arthritis with any of the followings: Palmoplantar pustulosis Pustular psoriasis Severe acne
Any severe osteitis** with any of the followings: Palmoplantar pustulosis Pustular psoriasis Severeacne

Any of three presentations is sufficient for the diagnosis

*Or with the presence of Propionibacterium acnes

**Involvement of a single site, including spondylodiscitis, is sufficient

Table 1 Diagnostic criteria for Synovitis-acne-pustulosis-hyperostosis-osteomyelitis(SAPHO) Syndrome

(Kahn MF et al.,1994)³⁾

	Healing	Improvement	No change
Skin lesion	5/12 cases	7	0
Bone & articular lesion	4/12 cases	7	1

Followed period 1yr-4yrs and 3month
(Ave.) 1yrs and 5months±12months

Table 2. Result by Dental Treatments

Treatment methods

Dental treatments (treatment for dental chronic focuses, dental metal removal by patch test and EPMA) for these cases was carried out.

In one case with wide spread chronic diffuse sclerosing osteomyelitis of the mandible with pathologic fracture, hemimandiblectomy, teeth extraction and the reconstruction for the resected hemi-mandible using a microvascular free flap of fibula were done. Tonsillectomy was done in only 2 cases.

Analyze methods for trace metals in the blood samples:

We took blood samples more than 2 hours after meals, before patch test for dental metals. The whole blood sample or separated serum samples were tested at SRL Co., Ltd. in Tokyo. The concentrations were analyzed colorimetry or atomic absorption analysis. The analyzed trace metals were serum zinc (Zn), serum copper (Cu), serum iron (Fe), blood selenium (Se) and blood mercury (Hg).

The Student's t-test was used for statistical analysis of the data.

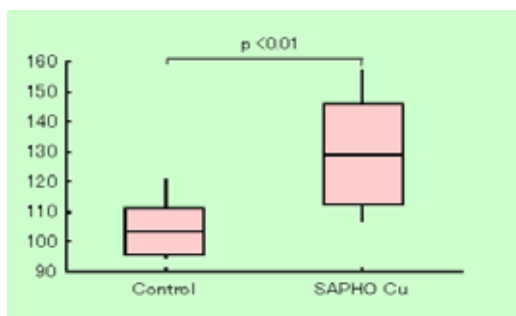


Fig. 1 The serum copper level

Results and discussion

The symptoms on bones and joints were as follows:

1 case of sternoclavicular-joint- hypertrophy, 10 cases of sternoclavicular-sternocostal-joint-pain, 4 cases in all had some other bones and joints pain. With regards to the oral aspects, all cases had dental chronic focuses, and 46% of cases had tonsillar chronic focus. There were dental-metal-restorations in 11 cases.

By the dental and surgical treatments, the skin lesion had been cured or had decreased remarkably in all cases, and pains in bones and joints had been cured in all cases.(Table 2)

We reported about clearly effect of dental treatment, removing focuses and dental metals, for cutis simptome of palmoplantar pustulosis. The rate of effect was more than 80% in 60 cases.²⁾ But we could not find a report about the effect of dental treatment for bone and joint symptoms of SAPHO syndrome with

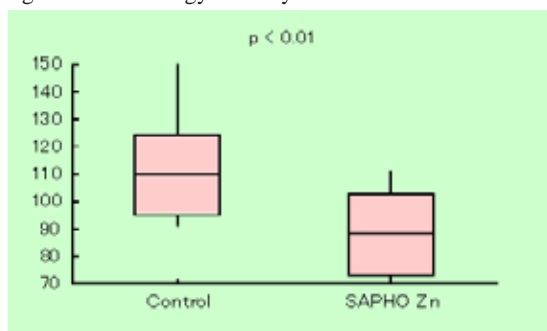


Fig.2 The serum zinc level

palmoplantar pustulosis. This study showed the effect of dental treatment clearly for not only skin but also bone and joint symptoms. In recent, a hypothesis "auto-immuno disease" was set up about pathogenesis of palmoplantar pustulosis and SAPHO syndrome. The clear effect of dental treatment for these diseases may suggest a possibility of relation between dental focus, dental metal and auto-immuno disease.

The results on the trace metals in the blood patient were as follows: The serum copper level was significantly high than healthy control(Fig.1). This result may reflect the activity of inflammation, as some reports suggested already. The serum zinc level was significantly low(Fig.2). Serum zinc is biomedical essential element, a part of roles were reported as antioxidative effect and immunopotentiative action. The low level zinc in serum may relate from immuno system. The blood selenium level was significant low (Fig.3) than healthy control. Blood selenium is biomedical essential element and was reported relation with immnosystem.too. The other trace metals in the blood showed no remarkable change.

These results about the essential trace metals may suggest a possibility that chronic lack or overdose of these metals, if it is a slight, have some relation to pathogenesis of SAPHO syndrome.

Conclusions

1. Dental treatment for SAPHO syndrome is very effective for skin and bone lesions.
2. Significant change of blood trace metals on the patient were found.
3. These result suggests a possibility intra-oral-focus, dental-metals and trace metals has some relation to pathogenesis of SAPHO syndrome.

References

- 1) Chamot AM, Benhamou CL, Kahn MF, Kaplan G: Prost A. Rev Rhum Mal Osteoartic, 54:187-196,1987
- 2) Ishiguro H, Mori K, Mataga I: Shigaku, 88: 256-271, 2000
- 3) Kahn MF, Kahn MS. Bailliere's Clinical Reumatology, 8: 333-362,1994