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Case Report

The efficacy of dental therapy for an adult case of Henoch–Schönlein purpura

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ABSTRACT

Henoch–Schönlein purpura (HSP) is a disease that involves palpable purpura on the skin, joint pain, and gastrointestinal problems. An acute bacterial infection is one of the causes of HSP. Odontogenic infectious diseases have also been implicated in causing HSP. However, there are only a few reports describing the correlation between HSP and odontogenic infectious diseases. The present study describes a case in which a patient was cured of HSP following dental treatment.

1. Introduction

Henoch–Schönlein purpura (HSP) is one of the vasculitis syndromes that affects small blood vessels. It is an acute, systemic, immune complex-mediated, leukocytoclastic vasculitis characterized by a triad of palpable purpura (without thrombocytopenia), abdominal pain, and arthritis. Most patients with HSP experience an antecedent upper respiratory illness. In patients with HSP, immunoglobulin A (IgA) immune complexes are deposited in small vessels, as a result of exposure to an antigen from an infection, medication, or other environmental factors. Group A streptococci (GAS), which can cause an upper respiratory tract infection, are the most common pathogenic microorganisms that cause HSP [1]. A majority of such cases occur in children, however it can also occur in adults. HSP is known to have a high probability of being spontaneously cured if supportive treatment is the primary intervention. However, it sometimes develops into severe conditions with a high rate of reoccurrence [1,2]. No form of therapy has ever been shown to decrease the duration of the disease or prevent recurrences. Nephritis is the most serious long-term complication of HSP. Although early aggressive therapy has been recommended for children and adults with such severe renal involvement, there is little evidence to indicate the best treatment for it [3].

Odontogenic infectious diseases are suspected to cause HSP. However, the relationship between HSP and odontogenic infectious diseases is unclear.

In the present study, we encountered a case which links HSP and odontogenic infectious diseases.

2. Case report

The time-line chart of the case history is shown in Fig. 1. A 39-year-old woman suffering from HSP, otherwise in good health, was referred to our department (oral and maxillofacial surgery) from the department of dermatology to see if dental treatment could improve her purpura in August 2010. She had developed swelling, pain, and purpura on her lower limbs which disappeared one week later in February 2010. After that, her symptoms reoccurred for several months. In July 2010, the frequency of the reoccurrences had increased, and the area of purpura extended to cover her thighs. She also occasionally experienced stomachaches. However, she had not experienced any joint pain or a history of fever. When she presented herself to the department of dermatology in our hospital, she underwent a biopsy. The results were positive for leukocytoclastic vasculitis, therefore suggesting HSP. A Lupus band test showed the deposition of IgA in dermal vessels and this result is consistent with HSP rather than cutaneous leucocytoclastic angiitis as a diagnosis. The Rumpel–Leede test was also performed and the result was negative. Laboratory data did not strongly indicate HSP with a serum IgA level of 301 mg/dl (normal) and factor XIII activity at 75.5% (normal). However, the platelet count was $15.6 \times 10^4/\mu l$ (normal), which eliminated the possibility of thrombocytopenic purpura and leukemia. Prothrombin time-international normalized ratio was 0.82 (not prolonged) and activated partial thromboplastin time was

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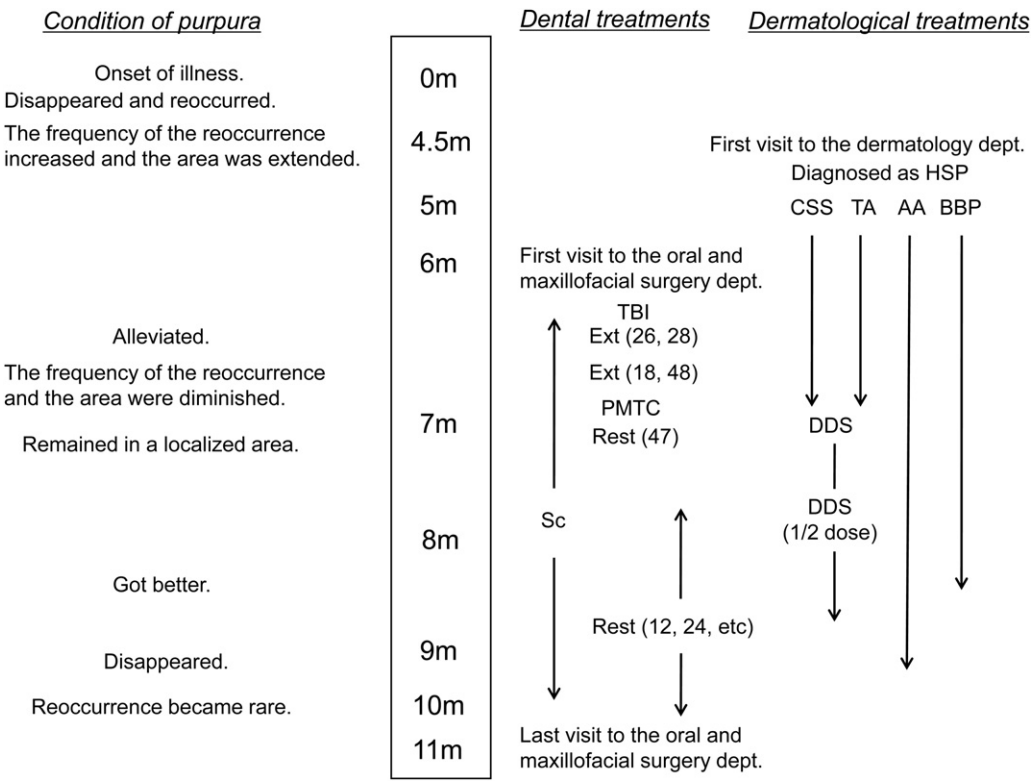

Fig. 1. The time-line chart of case history. A 39-year-old woman presented with recurrent painful purpura in the lower limbs from February 2010 (onset of illness, 0 m). Initially she was diagnosed at the department of dermatology as having Henoch–Schönlein purpura (HSP). There was mild stomachache but no arthritis. Shortly after being diagnosed she improved but purpura reoccurred and treatment with oral carbazochrome sodium sulfonate (CSS), tranexamic acid (TA), and ascorbic acid (AA), and also topical treatment with steroids, betamethasone butyrate propionate (BBP), was initiated (5 m). Simultaneously she was referred to the department of oral and maxillofacial surgery to evaluate whether dental treatment could improve her condition (6 m). Dental therapy toward oral infections included extraction of one infected tooth (26) as well as caries (28) and periodontal treatment was performed. Also previous oral medication with the exception of AA was stopped (7 m). The patient was instead prescribed oral diaphenylsulfone (DDS) during approximately one month and with a reduction of the daily dose during that period. The purpura gradually dissolved and finally disappeared along with the dental treatment and oral medication. m, month; TBI, tooth brushing instruction; Ext, extraction; Sc, scaling; rest, restoration; PMTC, professional mechanical tooth cleaning.

26.3 (not prolonged either), which excluded hemophilia. Gastro- and colonofiberscopy showed no abnormal findings related to HSP. Urinalysis showed hematuria only once. An analysis of bacterial cultures from the patient's nose found normal flora, *Neisseria* species, *Staphylococcus* species, α-streptococci, and methicillin-sensitive *Staphylococcus aureus*. Considering the results from all the tests and findings, HSP was the most appropriate diagnosis of her disease. About two weeks after the first visit to the department of dermatology, her purpura reoccurred, and carbazochrome sodium sulfonate (CSS, 30 mg), tranexamic acid (TA, 250 mg), and ascorbic acid (AA, 1000 mg) were prescribed to be taken after each meal. All of the medicine was meant to reinforce the wall of blood vessels. At the same time, the application of betamethasone butyrate propionate (BBP) ointment which has anti-inflammatory and immunosuppressive properties was started.

An odontogenic focal infection was suspected as a possible cause of her HSP because she had left dental treatments unfinished. She was referred to our department of oral and maxillofacial surgery and examined. An extraoral examination revealed no abnormalities except purpura on her lower limbs (Fig. 2). An intraoral examination revealed the deposition of plaque around the cervical region of almost all the teeth, calculus deposition on the proximal and cervical region of several teeth, many carious or restored teeth (18: caries; 16: full cast crown; 15: metal inlay; 14: metal inlay; 12: composite resin; 11: composite resin; 24: caries; 25: metal inlay; 26: caries endodontically treated before; 28: caries; 37: caries; 36: metal inlay; 44: resin veneered crown; 45: metal inlay; 46: metal inlay; 47: caries; and 48: caries), and halitosis. No mucosal lesions (tumor, ulcer, pus discharge, injury, etc.) were found (Fig. 3). X-rays showed little horizontal bone absorption and a radiolucent area around the apex of 26's palatal root. The radiopaque object in the distal buccal root of the same tooth was thought to be a reamer or a file (Fig. 4).

The course of action we decided to follow was periodontal treatment along with teeth extraction, caries restoration, and prosthodontic treatment. This would allow the lesions which might have been responsible for the focal infection to be effectively removed.

First, the plaque control record (PCR) and periodontal tissues were examined (Table 1) and tooth brushing instruction (TBI) was performed. Then the teeth that might have been responsible for HSP (26, 28) and useless teeth (18, 48) were extracted. Professional mechanical tooth cleaning (PMTC) and restoration procedures (12, 24, and 47) were then conducted.

Table 1
Periodontal risk assessment of the patient.

	Before treatment	After treatment
# of teeth	30	26
PCR (%)	60.7%	16.3%
# of teeth with PPD ≥ 4 mm	5	0
# of BOP-positive sites	21	8
# of mobile teeth	0	0

PCR, plaque control record; PPD, probing pocket depth; BOP, bleeding on probing.

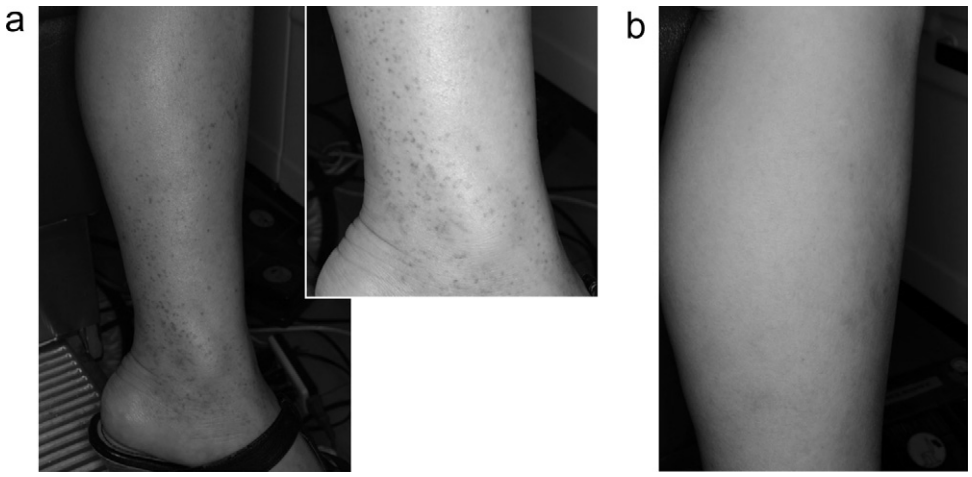

Fig. 2. (a) Purpura on the lower limbs on the first visit. (b) Lower limbs without purpura on the last visit.

The frequency and area of the patient's purpura were diminished subjectively during the dental treatment. One week after the extraction of 26, which had a periapical periodontitis, and 28, which was one of the carious teeth (Fig. 3), and TBI, the patient recognized that the purpura had been alleviated. After the extraction of 18 and 48, restoration of 47, and scaling, the purpura had got better, which reduced the frequency of the application of the ointment, but remained in a small and localized area. She was then instructed to finish taking the CSS and TA that had been prescribed by her dermatologist, to continue taking AA, and to take diaphenylsulfone (DDS, 25 mg) twice a day orally to reduce the inflammation of blood vessels. She continued to receive periodontal and restorative treatment in our department. About one month later, her purpura got better and then, the application of the ointment was stopped and dosage of DDS was reduced to once a day. After one and a half weeks of taking DDS, which finished her medication, the purpura disappeared and has reoccurred only once since then (Fig. 2).

The dental treatment was continued for about two and a half months after finishing the DDS. On the last visit, her PCR improved dramatically with the score of 16.3% (60.7% on the first examination). Probing pocket depth (PPD) showed little change since it was near normal on the first visit, but the number of bleeding on probing (BOP) sites decreased (Table 1).

3. Discussion

Several dermatological or autoimmune diseases are thought to correlate with odontogenic infectious diseases. For example, Burger's disease has been linked to periodontitis, but it has not been proven epidemiologically [2]. Palmoplantar pustulosis and chronic pigmented purpura have also been reported to have an association with an oral focal infection [4].

HSP is considered to be associated with odontogenic infectious diseases as well. However, there is little evidence as to the causal relationships and efficacy of dental treatment in easing HSP. There are a few reports that mention the correlation between HSP and odontogenic infectious diseases. Jinous et al. have reported on a case of HSP that had developed after endodontic treatment. This

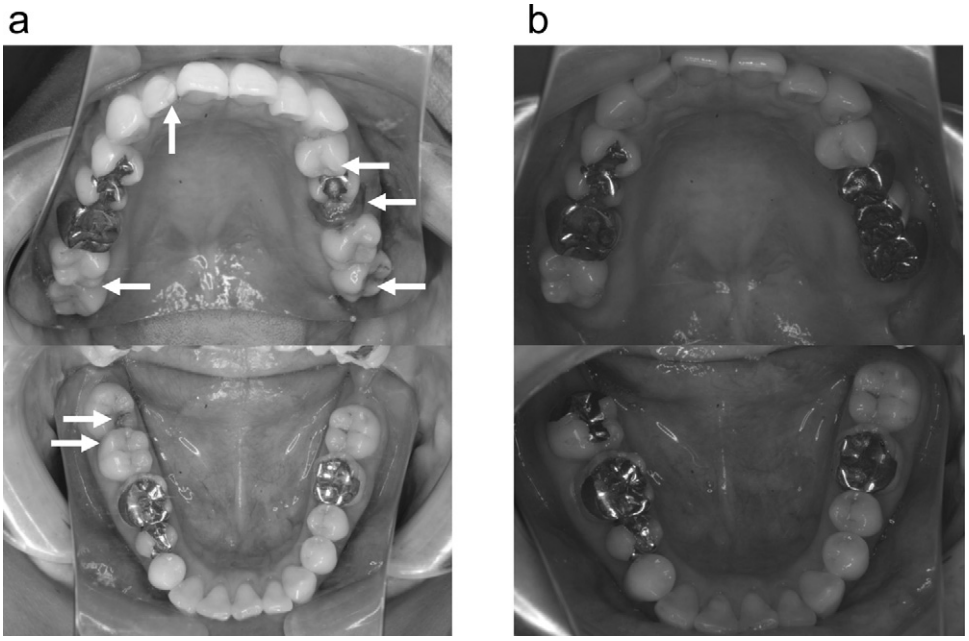

Fig. 3. (a) Oral photos before treatment. There were many carious teeth that needed to be treated and restored teeth (arrowheads). (b) Oral photos after the course of dental treatment. 18, 26, 28, and 48 were extracted. 12 and 24 were restored using light cured composite resin (CR). 47 was restored with a metal inlay. 26 was replaced with 25–27 metal bridge.

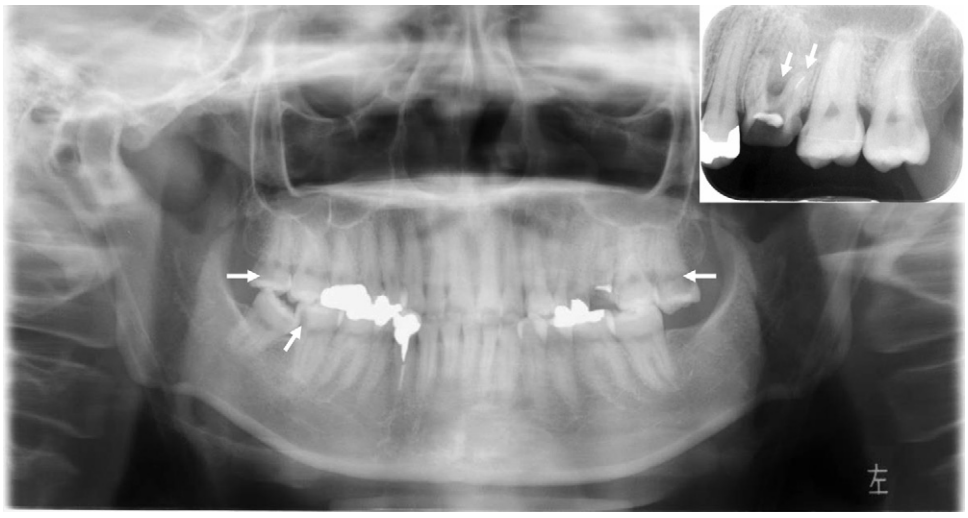

Fig. 4. Orthopantomographic and dental X-ray images of the first visit. There was a radiolucent lesion on the apex of the palatal root of 26, and a radiopaque object which was supposed to be a reamer or a file in the distal buccal root of the same tooth in the dental X-ray (upper right, arrowheads). There were many carious teeth and restored teeth. Remarkable carious teeth are indicated by arrowheads in the orthopantomographic X-ray. No significant horizontal or vertical alveolar bone absorption was found.

report suggested that root canal treatment could be a trigger for HSP [5]. The authors assumed that trepanation of the apex may cause a streptococcal bacteremia and that the change of the environment and microbiological flora of the root canal may cause a bacteremia. Inoue et al. have reported on the efficacy of dental treatment in preventing nephropathy in pediatric HSP. In their study, children with HSP underwent antimicrobial and dental, ear, nose, and throat treatment. Almost all the patients were cured by the treatments [6]. Igawa et al. reported that an oral focal infection could be a precipitating factor for adult HSP. Half of the patients having adult HSP presented with an oral focal infection and underwent dental treatment, including tooth extraction. A few weeks after tooth extraction, improvements in the skin lesions were observed [7].

These previous reports were described mainly by dermatologists, pediatricians, etc., and not by dentists. Therefore, the method of oral examination is considered to be different from that of dentists. For example, the evaluation of the teeth with a periapical lesion was performed by X-rays and other information such as the onset of periapical periodontitis or antecedent caries, types of pain, etc., were not mentioned. Furthermore, all of these reports were focused on periapical periodontitis as a common odontogenic infectious disease, and in this case only periapical periodontitis, but not "peripheral" periodontitis which is another common odontogenic infectious disease, was properly evaluated. It is said that if a person had 28 of their teeth with 4 or 5 mm of periodontal pockets, respectively, the potential infection sites would be summed up to be 50 or 72 cm^2 [8]. Every clinician needs to focus on "peripheral" periodontitis as another important odontogenic infectious disease which could be associated with HSP.

The patient we have taken care of in the present study had endodontic and periodontal problems, as well as dermatological symptoms. The patient was treated by means of tooth extraction, periodontal treatment, and medication. As a result, her purpura got better, but the factor that was most effective in treating her symptoms remains unclear. There was a relatively large periapical granuloma on the palatal root apex of 26. It is reasonable to regard 26 extraction as the most effective treatment. Our result suggests that periodontal infection, here periapical periodontitis, is the main candidate for the cause or maintaining factor of HSP in this case. The patient did not have any deeper periodontal pockets than 4 mm, but did have some BOP sites that could lead to HSP.

The medication regimen she had undergone was the same as that given to other patients. Therefore that might have been the common treatment which caused her HSP to improve. Furthermore, she might have gotten better without treatment. It has been reported that HSP is spontaneously cured in 89% of adult patients [1]. The patient felt that her symptoms had improved after the treatments in our office. However, this was difficult to discern since it was impossible to determine the frequency of purpura, to count the number of it, and to measure the area of it. Judging from these results, it is reasonable to say that the dental treatment improved her purpura, but it cannot be proved definitely.

To further understand the disease and to determine the cause of HSP, a proper study design and a large number of HSP patients with odontogenic infectious diseases are necessary.

In conclusion, we have experienced a case in which dental treatment may have led to improving purpura on the lower limbs of an adult HSP patient.

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References

[1] Reamy BV, Williams PM. Henoch–Schönlein purpura. *Am Fam Physician* 2009;80:697–704.
[2] Ozaki S, Ando M, Isobe M, et al. Guideline for management of vasculitis syndrome. *Circ J* 2011;75:474–503.
[3] Zaffanello M, Brugnara M, Franchini M. Therapy for children with Henoch–Schönlein purpura nephritis: a systematic review. *Sci World J* 2007;7:20–30.
[4] Satoh T, Yokozeki H, Nishioka K. Chronic pigmented purpura associated with odontogenic infection. *J Am Acad Dermatol* 2002;46:942–4.
[5] Tahmassebi JF, Paterson SA. Development of acute Henoch–Schönlein purpura subsequent to endodontic treatment. *Int J Paediatr Dent* 2007;17:217–22.
[6] Inoue CN, Nagasaka T, Matsutani S, et al. Efficacy of early dental and ENT therapy in preventing nephropathy in pediatric Henoch–Schönlein purpura. *Clin Rheumatol* 2008;27:1489–96.
[7] Igawa K, Satoh T, Yokozeki H. Possible association of Henoch–Schönlein purpura in adults with odontogenic focal infection. *Int J Dermatol* 2011;50:277–9.
[8] Hujoel PP, White BA, Garcia RI, et al. The dentogingival epithelial surface area revisited. *J Periodont Res* 2001;36:48–55.