DOI: 10.1111/j.1365-263X.2006.00810.x

Development of acute Henoch–Schönlein purpura subsequent to endodontic treatment

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International Journal of Paediatric Dentistry 2007; 17: 217–222

Background. Henoch–Schönlein purpura (HSP) is an inflammatory disorder of unknown origin that is characterized by IgA-dominant immune complexes in smaller blood vessels. It results in a triad of symptoms, including a purpuric rash on the lower extremities, abdominal pain or renal involvement, and arthritis. Any of the triad may be absent, however, which often leads to confusion in diagnosing the condition. Cases of acute HSP developing subsequent to dental treatment have not been previously reported in the dental literature.

Case report. This study reports the unusual case of a 14-year-old female who developed acute HSP following endodontic treatment.

Conclusion. Treatment for this condition is supportive and children affected by this disorder need close follow-up of their renal status.

Introduction

Henoch–Schönlein purpura (HSP) is a purpuric and systemic vasculitis of childhood[1]. It represents a diffuse vasculitis that is secondary to hypersensitivity, and occurs in twice as many males as females. The condition has been reported to have an incidence of 14 cases per 100 000 school-aged children, and appears to occur most frequently in the spring and autumn[2]. Over 75% of patients affected are aged between 2 and 11 years[3]. A recent prospective study estimated that the annual incidence of HSP in the UK is 20.4 per 100 000, and that the peak prevalence is in children aged between 3 and 10 years (the condition is rarely diagnosed in infants and young children). White Caucasians are more affected than black ethnic populations[4].

The most common symptoms of HSP include: purple spots on the skin (95–100% of cases), especially involving the legs; subcutaneous oedema (20–50% of cases); abdominal pain (85% of cases); and joint pain (60–80% of cases), especially involving knees and ankles. It can masquerade as many different conditions, depending on the symptoms.

Purpura may be defined as visible, unblanching haemorrhages in the skin, or mucous membranes that are 5–10 mm in diameter and often palpable[5]. The purpuric areas evolve from red to purple, become rust-coloured with a brownish hue and then fade[6]. In more severe cases, haemorrhagic, purpuric or necrotic lesions may be prominent. It is important to differentiate these lesions from those of meningococcal septicaemia or other septic emboli[7].

Although the cause of HSP is unknown, it is often associated with infectious agents such as group A streptococci and mycoplasma. It is also associated with food reactions, exposure to cold, insect bites and drug allergies[8]. Diagnosis of HSP depends on clinical findings and history. It is usually not difficult if the classic triad of rash, gastrointestinal complaints or haematuria, and arthritis is present. When symptoms are not typical, however, the differential diagnosis can become extensive (Table 1). There is not a specific laboratory test for the disorder, although an elevated serum IgA level is suggestive. Some laboratory studies can also help in excluding other diagnoses and in evaluating renal function, including urinalysis. Haematuria and/or proteinuria are present in 10–20% of patients[9].

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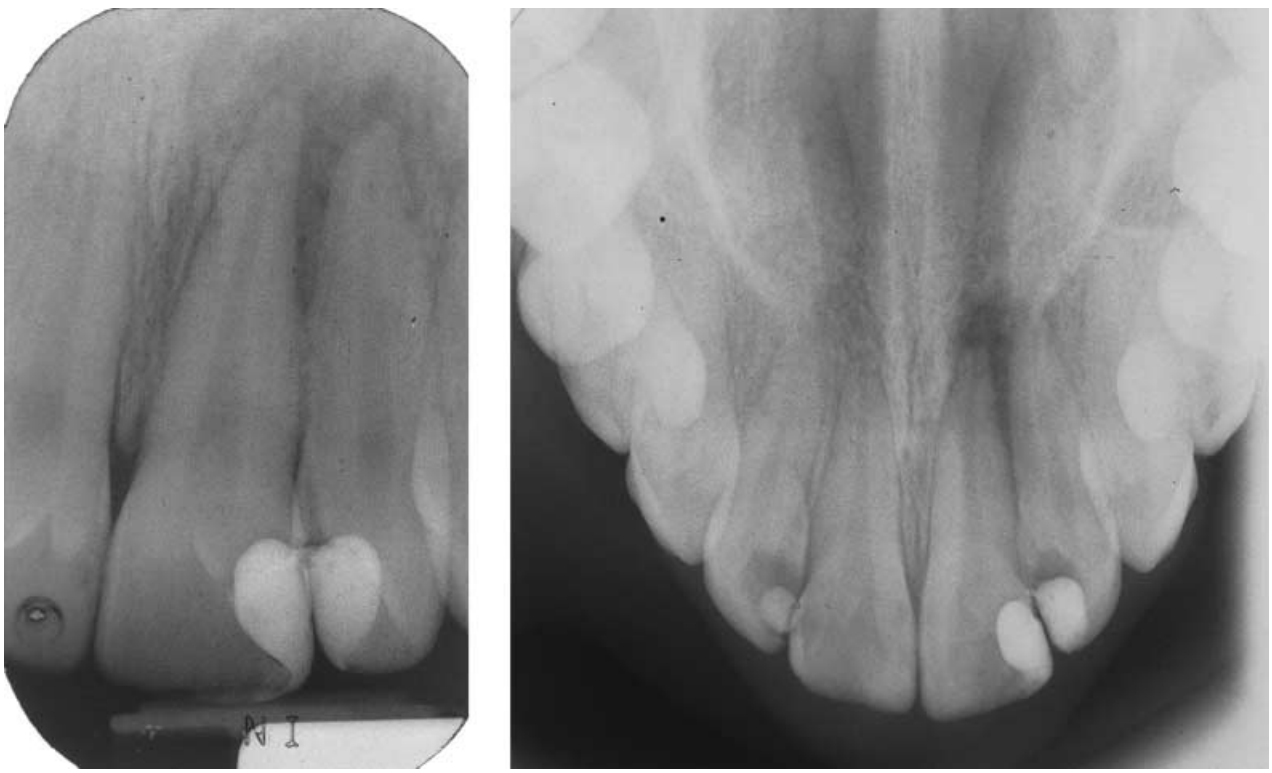
Table 1. Differential diagnosis of Henoch–Schönlein purpura.

Acute abdomen
Meningococcal meningitis or septicaemia
Rheumatoid arthritis
Systemic lupus erythematosus
Rheumatic fever
Idiopathic thrombocytopenic purpura
Drug reactions
Child abuse

Treatment of HSP is mostly supportive and includes ensuring adequate hydration and bed rest. Most cases resolve spontaneously without treatment. If symptoms persist non-steroidal anti-inflammatory drugs can relieve joint and soft-tissue discomfort. Clinicians often use corticosteroids to treat abdominal pain, subcutaneous oedema and nephritis. Treatment in patients with severe HSP nephritis includes many modalities; for example, intravenous or oral steroids with or without any of the following: azathioprine, cyclophosphamide and danazol[10]. In the absence of renal disease involvement, the prognosis for patients with HSP is excellent. The illness usually lasts from 4 to 6 weeks in most cases. Approximately 10–20% of patients have recurrences, and fewer than 5% of patients develop chronic HSP[11]. The following report describes the first reported case of acute HSP developing subsequent to dental treatment.

Case report

An anxious 14-year-old female was referred to the Leeds Dental Institute (LDI), Leeds, UK, for comprehensive dental care involving endodontic treatment of the upper left central and lateral incisors under inhalation sedation. There was no significant medical history reported. Extraoral examination revealed no abnormalities. Intraoral examination revealed no soft-tissue abnormalities except a discharging sinus on the upper left central incisor. Both the upper left central (UL1) and lateral incisors (UL2) were found to have large carious cavities; however, percussion and palpation were all within normal limits. Both periapical and occlusal radiographs of these teeth showed periapical periodontitis on the UL1 and UL2 (Figs 1 & 2).



Figs 1 & 2. Pre-operative periapical radiographs showing periapical periodontitis on the upper left central and lateral incisors.

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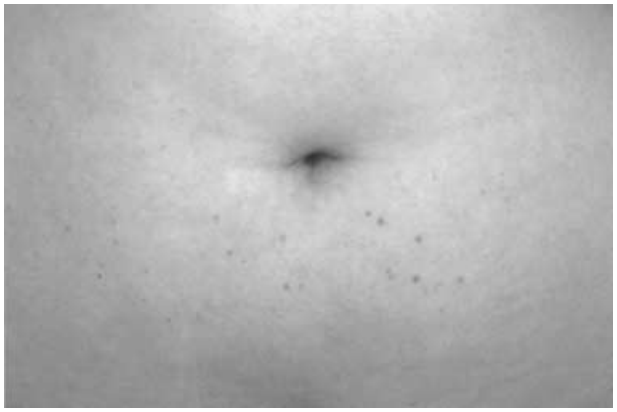
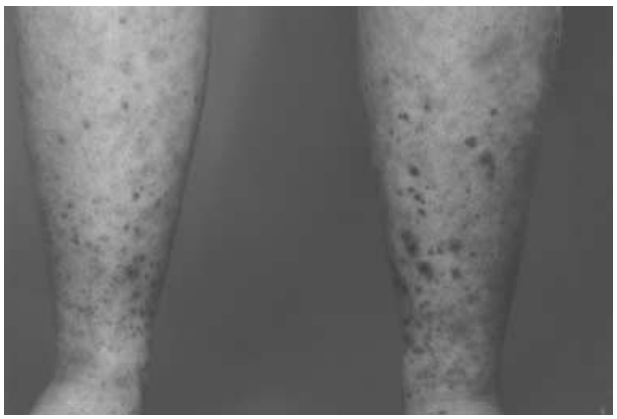
In January 2004, UL1 and UL2 were accessed under rubber dam using the crown-down chemotherapeutic technique, and the root canals were dressed with calcium hydroxide and glass ionomer dressing. The UL1 and UL2 incisor root canals remained symptomless, and UL1 was obturated in March 2004. The child's mother contacted the LDI 2 weeks after the obturation of UL1, informing the department that her daughter had developed swelling of her toes and legs, and a generalized rash one day after the last dental treatment. She had attended her general medical practitioner several times, and had been prescribed antibiotics and a steroid cream. No diagnosis had been made. The child's mother was concerned that her daughter's condition might have been related to the dental treatment that she had received. The patient was subsequently reviewed.

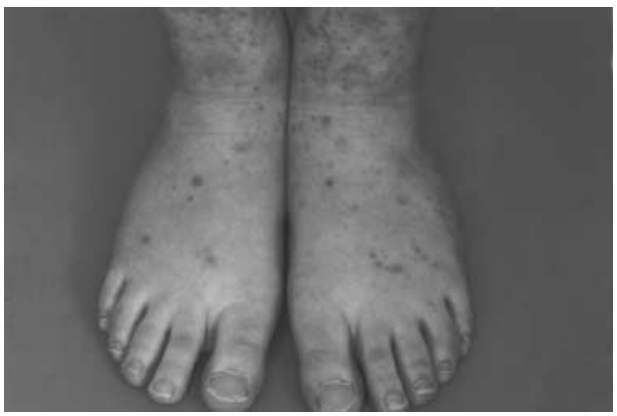
Extraorally, the patient had a purpuric rash on the arms, trunk, legs, feet and toes (Figs 3–5). Oedema of the legs, ankles and feet were present. Intraorally, UL1 and UL2 were symptomless. There was no presence of lymphadenopathy, no mobility of the UL1 and UL2, and there was no swelling or sinus present. A periapical radiograph showed a large radiolucent area associated with UL2 (Fig. 6). The patient had had root canal treatment of UL1 and UL2 carried out under inhalation sedation at separate visits because of her anxiety.

Microbiological analysis of UL2 was carried out in sterile conditions under rubber dam using an intracanal sterile paper point that was transported immediately to microbiology for culture and sensitivity. There was no pus exudate present in the canal; however, it appeared wet. Bacteria isolated from dentoalveolar infections include strict anaerobes, aerobes, microaerophillic and facultative bacteria (which include streptococci); however, the microbiology report only stated that streptococcal organisms were present in +++ amounts, and unfortunately, did not provide further detail.

An immediate referral was made to a paediatrician. Therefore, the child was admitted with a 5-week history of rash and swelling of her legs, abdomen, and to a lesser degree, her arms following the root canal treatment. A diagnosis of HSP nephritis was confirmed. The patient had persistent haematuria, proteinuria and glomerular nephritis with a blood pressure of 140/90. The child was taught to check urine using a dipstick, and was also advised to go on a low-salt diet and to elevate her legs when resting.







Figs 3, 4 & 5. Photographs showing purpuric rash present on the trunk, legs, feet and toes. Leg and ankle oedema is also visible.

The patient was reviewed 5 weeks after the original diagnosis by the paediatrician. The

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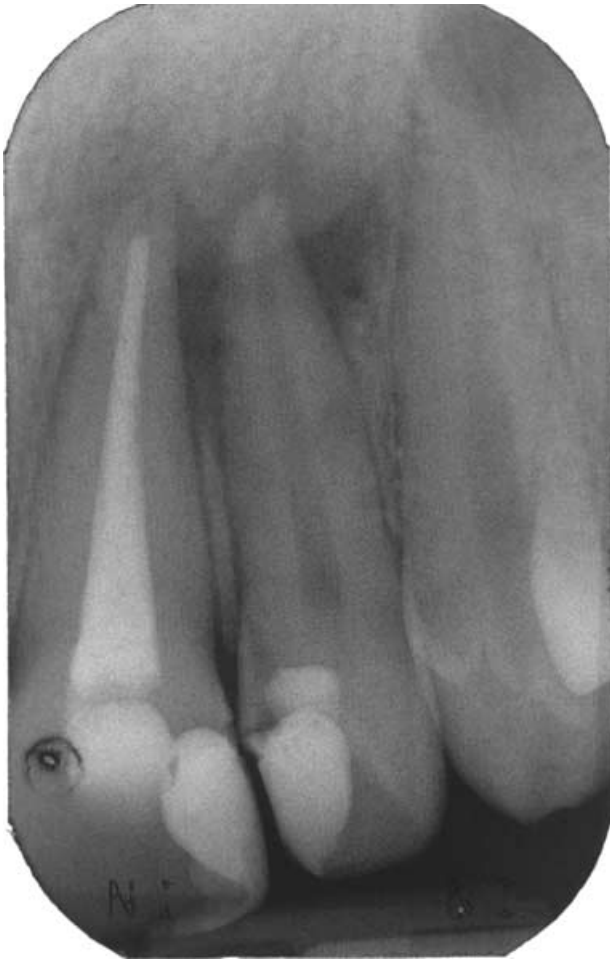
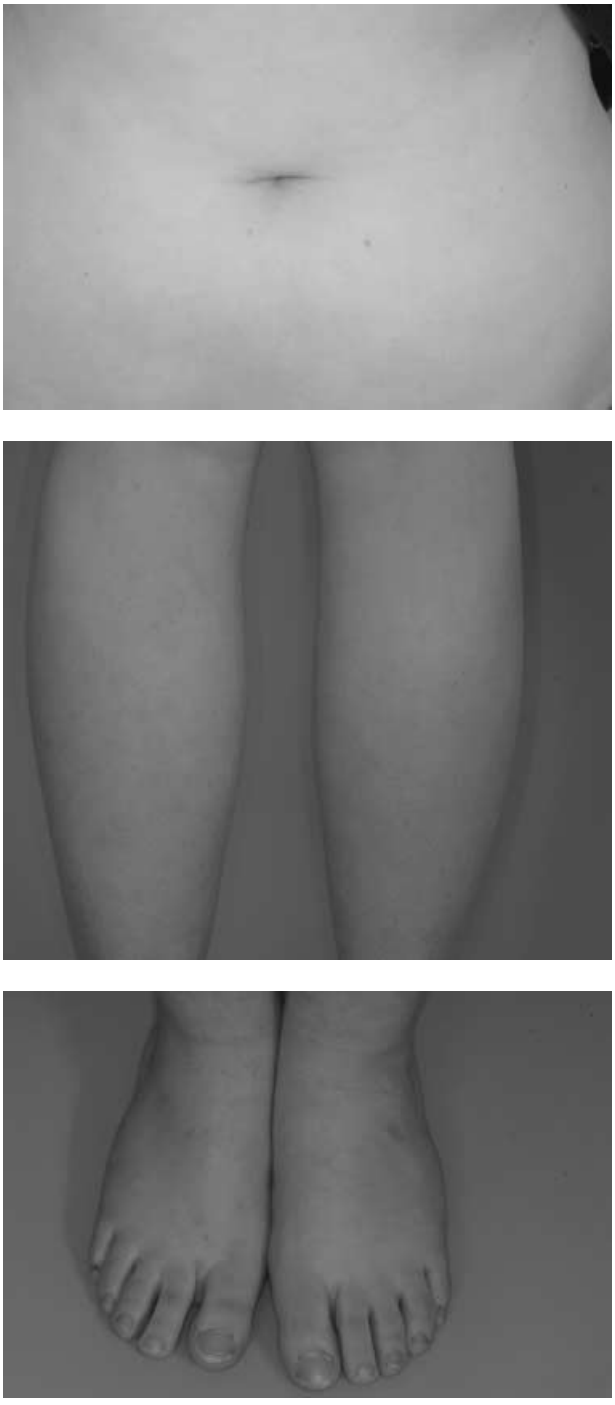


Fig. 6. Periapical radiograph showing a radiolucent area associated with the upper left lateral incisor.



Figs 7, 8 & 9. One-year follow-up photographs following the resolution of Henoch–Schönlein purpura showing no purpuric rash on the trunk, legs, feet or toes.

rash was found to be unchanged and there were still signs of microscopic haematuria with ongoing proteinuria, although the blood pressure had decreased to 137/67.

Subsequently, she was referred to the paediatric nephrologist with a view to carrying out a renal biopsy. When the patient was seen by the nephrologists approximately 12 weeks after the dental treatment, however, it was found that the patient's nephritis appeared to be settling and her urine protein/creatinine index was within normal limits. She was discharged, but was warned that the condition might reoccur.

After seeking advice from the consultant paediatrician, the root canal therapy was completed on UL2 in September 2004. The patient was reviewed one week after the root treatment, and there were no signs or symptoms of post-operative complications (Figs 7–9) and the acute HSP had resolved. Follow-up periapical radiographs of UL1 and UL2 in January 2006 showed good periapical healing (Fig. 10), and the patient reported no further

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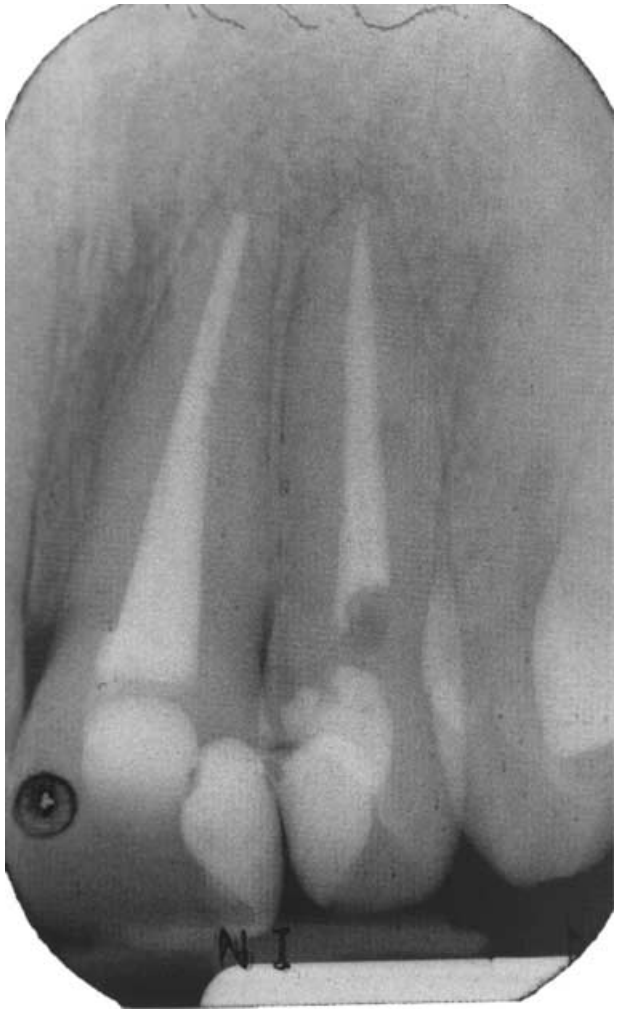

Fig. 10. One-year post-operative periapical radiograph showing periapical healing on the upper left central and lateral incisors.

episodes of HSP, although she has been kept under 6-monthly review by the paediatrician.

Discussion

Although HSP is a rare inflammatory disorder of childhood, it has clinical significance. The most serious sequela of HSP is renal involvement. This complication occurs in 50% of older children, but only 25% of children younger than 2 years of age. Fewer than 1% of cases progress to end-stage renal disease[12]. Patients who develop renal involvement generally do so within 3 months of the onset of rash. The most common manifestation of renal disease is haematuria. In the case reported here, the haematuria and proteinuria developed within 3 weeks of the rash. The presence of proteinuria and haematuria is associated with progression to renal insufficiency. In 50% of patients who display a combination of nephritis-nephrotic symptoms, end-stage disease develops after 10 years. In this case, the patient was seen by the nephrologists approximately 12 weeks after the dental treatment, and it was found that the patient's nephritis appeared to be settling and that her urine protein/creatinine index was within normal limits. She was discharged by the nephrologist, but was warned that the condition might reoccur.

Although HSP is generally a benign, self-limited condition, a close follow-up with repeated urinalysis is essential because a small percentage of patients progress to renal failure. This patient has been kept under 6-monthly review by the paediatrician.

A detailed history and examination of the patient did not reveal any known risk factors commonly associated with HSP, such as: infections (e.g. group A streptococci, mycoplasma and Epstein–Barr virus); vaccinations; or environmental exposures (e.g. drug and food allergens, cold exposure, and insect bites.) There were no known allergies and no latex allergy was present. The only apparent clinical risk factor to trigger HSP was the dentoalveolar infection associated with UL1 and UL2. There was a family history of HSP, which had been diagnosed in a cousin at less than 2 years of age. There may have been a familial susceptibility to HSP. In this patient's case, the dentoalveolar infection and periapical periodontitis were long-standing. It appears that the root canal treatment itself was the trigger for HSP, possibly because of trepanation of the apex, which may have caused a streptococcal bacteraemia, or by accessing the canal and carrying out the crown-down chemotherapeutic technique, root canal irrigants (e.g. sodium hypochlorite) may have altered the environment and microbiological flora of the canal, causing a bacteraemia. The appearance of a macular rash evolving into purpuric lesions occurred within a few days of obturation of UL1. The paediatrician confirmed that the aetiological trigger for HSP in this patient was either the root canal therapy or the long-standing dentoalveolar lesion.

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Because the incidence of HSP has increased in the UK population, the paediatric dentist needs to consider dental treatment and dental disease as potential triggers for HSP. Rashes in children are common; paediatric patients may develop a rash after prescription of antibiotics, especially penicillin or erythromycin, or hives associated with food allergies. It is important to exclude meningococcal meningitis, septicaemia and rheumatic fever in the differential diagnosis. Exclusion of HSP is essential in order to avoid renal complications. This case report also highlights the importance of accurate diagnosis in the provision of holistic care for paediatric dental patients.

What this paper adds

- This paper increases awareness and knowledge about HSP
- It also demonstrates that acute HSP can develop subsequent to dental treatment

Why this paper is important to paediatric dentists

- This paper is of value to paediatric dentists since, although HSP is generally a benign, self-limited condition, close follow-up with repeated urinalysis is essential because a small percentage of patients progress to renal failure

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