



Severe Periodontitis in a 5-year-old Girl With Hyperimmunoglobulin E Syndrome

P. Tsang, DMD G. Derkson, DMD R. Priddy, DDS, MSc A.K. Junker, MD
J. Slots, DDS, MS, PhD, DMD, MBA H. Larjava, DDS, PhD

Dr. Tsang is a pediatric dental resident, and Dr. Derkson is associate professor and head, Department of Dentistry, British Columbia's Children's Hospital, Vancouver, British Columbia, Canada; Dr. Priddy is director of Oral Medicine and Oral Pathology Postgraduate Hospital Residency Program, Department of Oral Biological & Medical Sciences, University of British Columbia; Dr. Junker is associate professor, Division of Infectious and Immunological Diseases, Department of Pediatrics, British Columbia's Children's Hospital and University of British Columbia; Dr. Slots is professor of periodontology and microbiology, School of Dentistry, University of Southern California, Los Angeles, Calif; Dr. Larjava is professor and director of Graduate Periodontics, chair of the Division of Periodontics and Dental Hygiene, Department of Oral Biological and Medical Sciences, University of British Columbia, Vancouver.
Correspond with Dr. Tsang at phoebetis@shaw.ca

Abstract

The hyperimmunoglobulin E syndrome (HIES) is a multisystem disorder that affects the: (1) dentition; (2) skeleton; (3) connective tissues; and (4) immune system. Little is known about periodontal manifestations of the syndrome. The purpose of this report was to describe a 5-year-old girl with suspected autosomal-recessive HIES syndrome who revealed profusely bleeding and painful gingiva and generalized aggressive periodontitis. A polymerase chain reaction (PCR)-based microbiological examination detected *Porphyromonas gingivalis*, *Tannerella forsythia*, *Prevotella nigrescens*, *Treponema denticola*, *Eikenella corrodens*, and *Campylobacter rectus* in the deep periodontitis lesions. The extraction of all deciduous teeth due to a poor prognosis and risk of systemic infection led to resolution of the oral inflammation. Long-term follow-up is required to determine the periodontal prognosis of the erupting permanent teeth. (*Pediatr Dent.* 2005;27:68-73)

KEYWORDS: HYPERIMMUNOGLOBULIN E SYNDROME, PERIODONTITIS, PRIMARY DENTITION

Received May 22, 2004 Revision Accepted November 2, 2004

Hyperimmunoglobulin E syndrome (HIES), also known as Job's syndrome or Buckley syndrome, is a primary immunological disorder of unknown etiology. HIES may debut with no genetic linkage, but autosomal-dominant and autosomal-recessive transmission patterns have been described.¹⁻⁶ The autosomal-dominant form of hyperimmunoglobulin E syndrome (AD-HIES) is commonly characterized by:

1. elevated serum immunoglobulin E levels;
2. chronic eczematoid dermatitis;
3. recurrent skin abscesses that often lack the usual signs of inflammation such as warmth, erythema, and tenderness ("cold abscesses");
4. lung staphylococcal infections with predisposition to formation of pneumatoceles;⁶⁻⁹
5. characteristic facies with facial asymmetry;
6. prominent forehead;
7. deep-set eyes;
8. broad nasal bridge;

9. mild prognathism;¹⁰
10. scoliosis;
11. joint hyperextensibility;
12. decreased bone density leading to a high risk of bone fracture after minimal trauma.¹¹

Retention of the primary teeth due to the lack of root resorption is a striking and unexplained feature which Grimbacher et al found in 72% of AD-HIES patients.¹ The autosomal-recessive form of HIES (AR-HIES) differs from AD-HIES in that:

1. skeletal and dental abnormalities are absent;
2. there is increased susceptibility to severe fungal and viral infections;
3. there is a high incidence of vascular and infectious central nervous system complications.¹²

The pathophysiology of the HIES syndrome remains unclear. Immune studies have attributed infection susceptibility to:

1. defective neutrophil chemotaxis⁶;
2. deficiency in humoral^{13,14} and cellular immune response^{8,15};

Table 1. Scoring System for Clinical and Laboratory Findings in HIES Patients*† Clinical Findings

Clinical Findings	Points									
	0	1	2	3	4	5	6	7	8	10
Highest serum-IgE level (IU/ml) [‡]	200	200-500			501-1000				1001-2000	>2000
Skin abscesses	None		1-2		3-4				>4	
Pneumonia (episodes over lifetime)	None		1		2		3		>3	
Parenchymal lung anomalies	Absent						Bronchiectasis		Pneumatocele	
Retained primary teeth	None	1	2		3				>3	
Scoliosis, maximum curvature	<10°		10°-14°		15°-20°				>20°	
Fractures with minor trauma	None				1-2				>2	
Highest eosinophil count (cells/ml)	<700			700-800			>800			
Characteristic face	Absent		Mildly present			Present				
Midline anomaly	Absent					Present				
Newborn rash	Absent				Present					
Eczema (worst stage)	Absent	Mild	Moderate		Severe					
Upper respiratory infections per year	1-2	3	4-6		>6					
Candidiasis	None	Oral	Fingernail		Systemic					
Other serious infections	None				Severe					
Fatal infection	Absent					Present				
Hyperextensibility	Absent					Present				
Lymphoma	Absent					Present				
Increased nasal width	<1 SD	1-2 SD		>2SD						
High palate	Absent		Present							
Young-age correction	15 years			2-5 yrs		1-2 yrs		<1 yr		

*Adapted from Grimbacher et al.¹

†Points are assigned to each finding on the basis of its incidence in and specificity for HIES. Based on the frequency and severity of HIES characteristics exhibited, HIES is considered highly likely, with an HIES score >40 points, and possible with a HIES score >20 points. At 10 to 15 points, the presence of HIES genotype is undetermined. At <10 points, the patient is unlikely to have HIES. Shaded area denotes the scores for the patient in this case, totaling 64 points.

‡Normal=<130 IU/ml.

3. dysregulation of T-cell cytokine signals.^{16,17}

The HIES diagnosis is based on the constellation of patients' clinical and laboratory features, as no specific diagnostic test is available. A HIES scoring system developed at the National Institutes of Health (NIH) to phenotype patients with AD-HIES is helpful (Table 1).¹ HIES management is dependent on:

1. prophylactic antibiotics;
2. local debridement;
3. surgical incision and drainage of infectious lesions.⁹

Only a limited number of articles have described oral findings in HIES patients. These can include oral ulcerations,¹⁸ gingivitis,¹⁸ and prolonged oral and cervicofacial infections.^{19,20} O'Connell et al found that 81% (13/16) of HIES-affected children 7 to 17 years old demonstrated a delay in permanent teeth eruption that amounted to more than standard deviation of the average eruption age. Twenty-eight percent (5/18) of HIES patients older than 20 years showed radiographic evidence of delayed exfoliation of primary teeth. Histological examination of retained deciduous teeth revealed an abnormal persistence of the Hertwig's epithelial root sheath, a finding that may partly be related to the primary teeth's delayed exfoliation.²¹

Since no previous report has described periodontitis in young HIES patients, the purpose of this report was to detail a 5-year-old girl with probable AR-HIES who exhibited advanced periodontitis in her entire primary dentition.

Case report

The 5-year-old female patient was the firstborn child of a Kurdish couple who are first cousins. She has 2 healthy younger brothers. None of her immediate family members

have problems with infections or other HIES features. She was born at full-term by spontaneous vaginal delivery after an uncomplicated pregnancy. Shortly after birth, she developed severe eczema which has been difficult to manage because of frequent *Staphylococcus aureus* and *Candida albicans* supra-infections.

She had recurrent otitis media. By age 6, she had been hospitalized with pneumonia on 5 occasions and had 1 episode of *S. aureus* sepsis. At age 3, she developed severe HSV1-positive gingivostomatitis, for which she was admitted to the hospital and treated with intravenous acyclovir. She had an uneventful course of chickenpox at age 2 1/2, but developed shingles at age 6. She was almost exclusively breast-fed until age 15 months. After weaning, she fed poorly on a restricted diet, which resulted in severe failure to thrive. She had osteopenia and suffered 2 tibial fractures after minor trauma.

With dietary supplements and nasogastric feeding of an elemental formula, her growth velocity normalized, but her height and weight remain below the third percentiles. Other clinical and laboratory findings (Table 1) resulted in a HIES score of 64. Without including the finding of osteopenia and fractures, considered at this time to be related to dietary insufficiency, her HIES score is 60. Definite diagnosis, however, is controversial and remains to be confirmed by future development of reliable molecular diagnostic markers.

At age 5, the patient was referred to the dental clinic at the British Columbia's Children's Hospital, Vancouver, British Columbia, Canada, with the chief complaint of painful gingival enlargements over the past 2 months. The mother reported significant gingival bleeding during brushing. The

Table 2. Summary of Oral Findings in a 5-year-old Girl With HIES Syndrome

Summary of Oral findings										
Extraoral examination										
<ul style="list-style-type: none"> • Dry cracked lips • Eczematous, scaly lesions on her cheeks 										
Intraoral examination										
<ul style="list-style-type: none"> • Smooth tongue with deep fissure • Malodor 										
Periodontal examination										
Gingiva										
<ul style="list-style-type: none"> • Red, edematous, rolled margin and blunted papillary • Boggy with loss of surface stippling • 100% bleeding on probing • Probing depth in mm at 6 sites per tooth (mesiobuccal, midbuccal, distobuccal, distolingual, midlingual, mesiolingual): 										
	Distal ←----- Mesial					Mesial -----→ Distal				
Buccal	6 6 6	6 6 6	6 6 6	5 5 3	3 3 3	3 3 3	3 3 3	6 3 4	5 3 6	6 6 6
Tooth	A	B	C	D	E	F	G	H	I	J
Lingual	5 5 5	6 4 6	6 3 4	4 3 3	3 3 3	3 3 3	3 3 3	4 3 6	6 3 7	7 3 3
Lingual	7 6 8	7 6 7	8 8 8	9 9 9	9 9 9	9 9 9	9 9 10	9 6 6	8 7 8	8 8 8
Tooth	T	S	R	Q	P	O	N	M	L	K
Buccal	6 6 6	6 6 6	9 9 12	9 9 9	9 9 9	9 9 9	9 9 9	9 6 6	9 6 6	6 9 6
	Distal ←----- Mesial					Mesial -----→ Distal				
Deposit										
<ul style="list-style-type: none"> • Minimal plaque deposit 										
Mobility:										
<ul style="list-style-type: none"> • Class III mobility on all teeth 										
Radiographic examination										
<ul style="list-style-type: none"> • Extensive horizontal alveolar bone loss 										

mother recalled no recent changes in medications or oral care. The extraoral examination revealed very dry, crusted lips and bilaterally erythematous cheeks. Her neck and extremities showed eczematous, excoriated scaling lesions. The intraoral findings are summarized in Table 2. Also, the tongue surface was smooth but deeply fissured. Gingiva appeared fiercely red and edematous with spontaneous bleeding (Figure 1). The child was in the primary dentition stage, and all teeth except nos. E, F, and G showed probing pocket depths of 5 mm or more and Class III mobility. Radiographic examination revealed advanced horizontal alveolar bone loss around all primary teeth (Figure 2). Minor interproximal dental caries lesions were detected.

For bacteriological examination, paper-point samples were collected from 4 advanced periodontitis lesions and processed separately. A polymerase chain reaction methodology was used to identify suspected periodontopathic species.²² The study lesions yielded *Porphyromonas gingivalis*, *Tannerella forsythia*, *Treponema denticola*, *Eikenella corrodens*, *Campylobacter*

rectus and *Prevotella nigrescens*. No sample revealed *Actinobacillus actinomycetemcomitans*.

The patient's advanced periodontitis prevented a conservative therapy. All primary teeth were extracted under general anaesthesia. Healing proceeded uneventfully and resulted in the elimination of soft tissue inflammation. The treatment aimed at:

1. minimizing the risk of infection by periodontopathic bacteria in the gingival crevice of erupting permanent teeth;
2. preventing a systemic dissemination of pathogens having the potential to cause pneumonia.

Histopathological analysis performed on 4 representative gingival lesions and associated teeth revealed heavily inflamed gingiva with elongated rete ridges and a predominance of plasma cells and intraepithelial polymorphonuclear leukocytes (Figures 3A to 3C). The root cementum appeared necrotic with no periodontal ligament attachment (not shown). The teeth showed no evidence of pulpal pathosis.



Figure 1. Red and erythematous gingiva with spontaneous bleeding in a 5-year-old girl with HIES syndrome.

The patient was re-evaluated at 1 and 6 weeks after the extractions. Her mother noticed a marked reduction in the child's oral discomfort and significant improvement in her appetite. Gingival healing was satisfactory, but regeneration of alveolar bone was not observed. Whether a period of edentulousness may prevent the recurrence of periodontitis in the permanent dentition remains to be determined with future monitoring. Prosthetic replacement of primary teeth and interceptive orthodontic treatment due to early loss of primary teeth are pending upon improvement of the patient's periodontal health.

Discussion

This child has clinical and laboratory features consistent with HIES. Having unaffected, consanguineous parents suggests the autosomal-recessive form of HIES. Her NIH-HIES score of 64 is in the 36 to 53 range that Renner et al determined in AR-HIES patients greater than age 1. Moreover, she has experienced problems with viral infections (herpes gingivostomatitis, shingles) that are common in this condition. Although other reports of oral complications in HIES mention oral ulcerations and gingivitis,¹⁸ the authors believe this is the first known report of generalized advanced periodontitis. Aggressive periodontitis around every tooth in a dentition is a very rare occurrence in prepubertal children. Considering the predisposition of HIES patients to recurrent infections, it is not surprising that such individuals are at risk of developing aggressive periodontal disease.

The periodontitis lesions studied yielded mixtures of *P gingivalis*, *T forsythia*, *T denticola*, *E corrodens*, *C rectus*, and *P nigrescens*. These bacteria are typically found in adults²³ and children²⁴ with severe periodontitis and gingivitis, but are usually not prominent in young children.²⁴ HIES-associated immunodeficiencies could permit periodontopathic bacteria to colonize young individuals. A *actinomycetemcomitans* was

not detected in the periodontitis lesions studied, even though the organism is a major pathogen in various types of destructive periodontal disease in children and adolescents.²⁴ In contrast to the present patient's periodontal condition, however, *A actinomycetemcomitans* is typically associated with a localized pattern of periodontal destruction and relatively little gingivitis.

HIES patients may experience an aggravated course of periodontitis because of defective polymorphonuclear leukocytes,⁶ deficient antibody responses,^{13,14} and changes in the T-helper (Th)1/Th2 balance towards a Th2 response.^{25,26} The Th2 predominance may accelerate periodontal breakdown through an overproduction of IgE.²⁷ The Th2-related interleukin (IL)-4 and IL-13 cytokines enhance IgE production, whereas the Th1-related interferon- γ and IL-12 suppress IgE production. The elevated IgE level, resulting from the Th2 predominance and reduced interferon- γ level,^{16,17} may cause a release of bone-resorbing prostaglandin-E₂,²⁸ IL-1 β , and tumor necrosis factor- α from monocytic cells.²⁹ At the same time, interferon- γ and transforming growth factor- β , which are major anti-inflammatory and bone resorption-inhibitory mediators, are reduced in HIES patients.²⁶ All together, rapid periodontal tissue destruction in HIES patients could be due to the combined effect of highly virulent periodontopathic bacteria, deficient polymorphonuclear leukocyte responses, increases in potent bone-resorbing cytokines, and decreases in bone resorption-inhibitory cytokines.

The ideal management of HIES-associated periodontitis would be to correct the underlying defects. HIES patients have been treated with histamine receptor 2 antagonist, cromoglycate, levamisole, isotretinoin, cyclosporine A, interferon- γ/α , and intravenous γ -globulin—all with limited effectiveness.³⁰ Bone marrow transplantation to correct the underlying immunodeficiency has failed to yield clinical improvement.³¹ The main management strategies continue to be: (1) prophylactic antibiotics; (2) timely treatment of infections; and (3) surgical intervention as necessary.

In terms of periodontal care, no study is available to delineate the extent to which HIES patients respond to conventional anti-infective treatment. Even though Papillon-Lefèvre syndrome has different pathogenic mechanisms, the management approaches for periodontitis in Papillon-Lefèvre syndrome patients may also be applicable to the severe periodontitis in this case. Treatment studies of Papillon-Lefèvre syndrome periodontitis show that extraction of hopeless teeth or the entire primary dentition decreases the risk of infection around later erupting teeth.^{32,33} All primary teeth were extracted in this patient. Whether or not a period of edentulousness improves the prognosis of the permanent teeth is unknown.

Conclusions

It should be realized that valuable diagnostic clues of a serious medical disorder might be obtained by identifying severe periodontal destruction along with salient systemic disease

characteristics. The cooperation between pediatricians and pediatric dentists is necessary for providing comprehensive treatment of children with systemic diseases.

Hyperimmunoglobulin E syndrome's clinicopathologic features are not uniformly expressed in individuals with the different genetic variants. The classic features include: (1) recurrent skin and lung staphylococcal infection; (2) chronic eczematoid dermatitis; and (3) elevated serum IgE level. Aggressive periodontitis may also be an associated phenotype. Efficacious periodontal therapy of HIES-related periodontitis remains to be determined.



Figure 2. Panoramic radiograph showing profound horizontal bone loss around all primary teeth in a 5-year-old girl with HIES syndrome.

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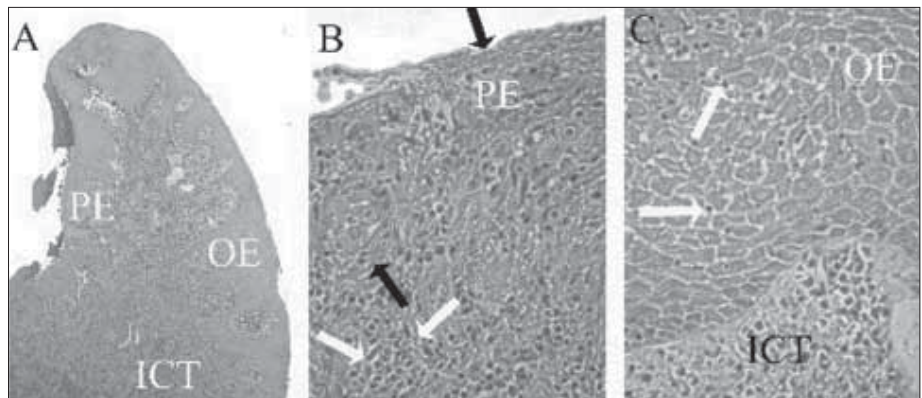


Figure 3A-C. Biopsy of inflamed gingival tissue from a 5-year-old girl with HIES syndrome. A) heavily inflamed gingiva with elongated rete ridges of the pocket epithelium and presence of predominantly plasma cells and intraepithelial polymorphonuclear leukocytes; B) higher magnification view of the pocket epithelium (between black arrows) and inflamed connective tissue (between white arrows); C) higher magnification view of oral epithelium infiltrated by polymorphonuclear leukocytes (white arrows). OE - Oral epithelium; PE - Pocket epithelium; ICT - Inflamed connective tissue.

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ABSTRACT OF THE SCIENTIFIC LITERATURE



SPONTANEOUS MANDIBULAR ARCH RESPONSE AFTER RAPID PALATAL EXPANSION

The purpose of this retrospective longitudinal study was to examine the spontaneous mandibular arch dimension changes in patients with class I malocclusions following rapid palatal expansion. Serial models of 30 patients with transverse discrepancies treated only with rapid palatal expansion in the early or mid-mixed dentition were measured for changes in: (1) mandibular arch width; (2) arch length; and (3) arch perimeter. Measurements were obtained at 4 assessment stages: (1) pre-expansion; (2) short-term follow-up; (3) progress; and (4) long-term follow-up. The authors reported a statistically significant increase in intermolar arch width following RPE, but reported no changes in: (1) intercanine width; (2) arch length; or (3) arch perimeter attributable to RPE.

Comments: This is one of those classic reports where the results are shown to be statistically significant, but the clinical significance is questionable. From pre-expansion to long-term follow-up (approximately 10 years), a mean increase in intermolar arch width of slightly less than 1 mm was observed ($P < .05$). **ALS**

Address correspondence to Dr. Anna Carolina Lima, Avenida Alberto Andaló, 4025, São José do Rio Preto, SP 15015-000, Brazil.

Lima AC, Andaló AA. Spontaneous mandibular arch response after rapid palatal expansion: A long-term study on class I malocclusion. *Am J Orthod Dentofacial Orthop.* 2004;126:576-582.

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