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ABOUT COVER

Editorial board member of *World Journal of Clinical Cases*, Dr. Kvolik is a Professor in the School of Medicine, Osijek University, Croatia. She obtained her MD degree, with specialization in the field of anesthesiology, resuscitation and intensive care from the Zagreb Medical School, Croatia. Afterwards, she undertook postgraduate training in Clinical Pharmacology at the same institution, defending both a Master's thesis and PhD thesis. In 2006, she joined the Osijek University Medical Faculty as a lecturer and was promoted to Professor in 2009. In 2012, she was elected Head of the Department of Anesthesiology, Resuscitation, Intensive Care and Pain Therapy, a position she occupies to this day. She is also the current Head of the Intensive Care Unit at the Osijek University Hospital, Croatia. (L-Editor: Filipodia)

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CASE REPORT

Acute generalized exanthematous pustulosis with airway mucosa involvement: A case report

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Author contributions: Li LL was involved in data curation, provision of resources, investigation and writing of the original draft; Li T performed data curation and formal analysis, participated in the investigation, project administration, methodology design, software utilization, data visualization, writing, reviewing and editing of the manuscript; Lu YQ took part in the provision of resources, supervision of the study, reviewing and editing of the manuscript; all authors have read and approved the final manuscript.

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Abstract

BACKGROUND

Acute generalized exanthematous pustulosis (AGEP) is a severe cutaneous adverse reaction characterized by sterile pustules on erythematous skin associated with fever and leukocytosis. The annual incidence of AGEP is estimated to be 1-5 cases per million. Cases of AGEP with oral mucosa involvement have been reported. However, reports of AGEP involving airway mucosa are limited.

CASE SUMMARY

We report a 42-year-old woman with serious AGEP involving the airway mucosa. The patient initially developed fever and a small rash on her forehead and face. Over the next 2 d, she developed a diffuse, pustular rash over her trunk and legs. In addition, she complained of a cough with white foam-like sputum, chest tightness and dyspnea. Four days later, due to dyspnea, her mental status started to gradually deteriorate. She became more and more drowsy. Biopsies of the skin and airway mucosa suggested the diagnosis of AGEP. According to the European study of severe cutaneous adverse reactions group's scoring system, the patient scored +6 indicating a probable diagnosis of AGEP. She received intravenous methylprednisolone 120 mg/12 h for 3 d, and was eventually discharged in good condition. This patient had already experienced respiratory failure and airway mucosa involvement on admission; however, the clinicians had an insufficient understanding of AGEP. Glucocorticoid was administered for more than 10 d following onset of the disease, and her overall prognosis was satisfactory.

CONCLUSION

This case represents a rare clinical feature of AGEP and an important finding for clinicians.

Key words: Acute generalized exanthematous pustulosis; Airway mucosa; Traditional Chinese medicine; Heavy metals; Case report

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Core tip: Acute generalized exanthematous pustulosis (AGEP) is a rare dermatologic reaction characterized by an erythematous rash with fever, leukocytosis and pustular erosions. We report a 42-year-old Chinese woman who developed serious AGEP involving the airway mucosa. Following treatment with intravenous methylprednisolone, the patient was eventually discharged in good condition. This case represents a rare clinical feature of AGEP and an important finding for clinicians.

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INTRODUCTION

Acute generalized exanthematous pustulosis (AGEP) is a severe cutaneous adverse reaction characterized by sterile pustules on erythematous skin associated with fever, leukocytosis and pustular erosions. The annual incidence of AGEP is estimated to be 1-5 cases per million[1,2]. It was first reported and named by Beyrot *et al*[3] in 1980. We present the rare case of a 42-year-old woman who developed serious AGEP involving the airway mucosa.

CASE PRESENTATION

Chief complaints

Rash, fever for 8 d, and dyspnea for 12 h.

History of present illness

The 42-year-old Chinese woman initially presented with fever and a small rash on her forehead and face. Over the next 2 d, she developed a diffuse, pustular, itchy rash over her trunk and legs. She also complained of a cough with white foam-like sputum, and chest tightness. Four days later, due to dyspnea, her mental status started to gradually deteriorate. She required supplemental oxygen. She then became more and more drowsy. Six days later, she received endotracheal intubation and ventilator support and was admitted to the intensive care unit. She was a housewife, 1.62 m in height with a body weight of 55.4 kg.

History of past illness

She had a history of lumbar disc herniation. She had taken traditional Chinese medicine for back pain 1 d before the onset of fever and rash.

Personal and family history

She had no other past medical history, and no family history of similar diseases or psoriasis.

Physical examination

Physical examination showed diffuse, pustular rashes over her trunk and extremities (Figure 1).

Laboratory examinations

Laboratory studies showed a white cell count of 15.3 × 10^9/L, neutrophils of 87.3%, C-reactive protein of 208.00 mg/L, and procalcitonin of 0.18 ng/mL. Blood gas analysis showed pH 7.35, PaO_2 62.1 mmHg, $PaCO_2$ 58.3 mmHg, and HCO_3^- 31 mmol/L. A large number of white cells were seen in the pustular fluid smear. Pustular fluid culture was negative. Anti-nuclear antibody, anti-neutrophil cytoplasm antibody, anti-mitochondrial antibody, TB-GeneXpert, and T-spot were within normal limits. All blood and pustular fluid cultures were negative.

Figure 1 Diffuse pustular rash of the trunk and legs. A: Diffuse, pustular rash of the trunk; B: Diffuse pustular rash of the legs.

Imaging examinations

Lung computed tomography showed that the airway space was occupied and tracheoscopy was recommended. Tracheoscopy revealed an unidentified protrusion in the main bronchus (Figure 2).

Pathological findings

Skin and airway mucosa biopsies showed subepidermal bullous formation containing scattered neutrophils and eosinophils with occasional subcorneal neutrophilic pustules, which suggested the diagnosis of AGEP (Figure 3).

FINAL DIAGNOSIS

According to the European study of severe cutaneous adverse reactions group scoring system, used in the identification of AGEP, the patient scored +6, indicating a probable diagnosis of AGEP. Her final diagnosis was AGEP, acute respiratory failure (type II), and pulmonary encephalopathy.

TREATMENT

On hospital day 5, broad spectrum antibiotics were discontinued. The patient then received intravenous methylprednisolone 120 mg/12 h on hospital day 6.

OUTCOME AND FOLLOW-UP

Following treatment with intravenous methylprednisolone, the patient's rash began to subside and there was desquamation of the skin after AGEP resolution (Figure 4A). The pustules gradually dried up and scabbed (Figure 4B). Post-treatment bronchoscopy showed that the airway was unobstructed (Figure 4). The patient was eventually discharged in good condition under instructions to avoid traditional Chinese medicine.

DISCUSSION

The diagnosis of AGEP includes the following: (1) Fever > 38°C; (2) Acute pustular eruption; (3) Neutrophilia; (4) Subcorneal or intra-epidermal pustules on skin biopsy; and (5) Spontaneous resolution within 15 d[1]. Our patient met all these features.

Cases of AGEP involving oral mucosa have been reported. However, cases of airway mucosa involvement are rare. The etiology of AGEP is not fully understood. More than 90% of AGEP cases are caused by drugs, such as aminopenicillins,

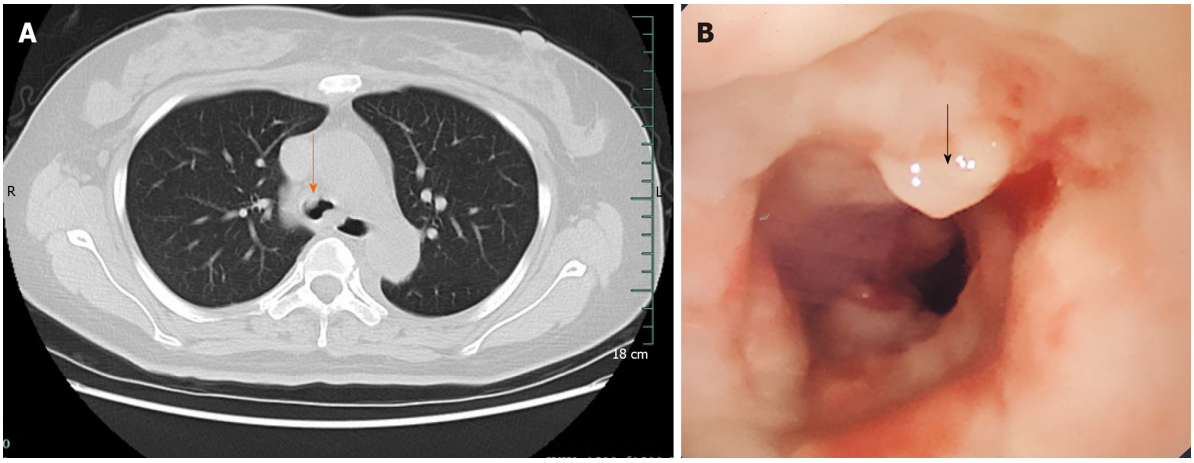

Figure 2 Lung computed tomography findings and the first tracheoscopy. A: Lung computed tomography; B: Tracheoscopy. The main bronchus was occupied.

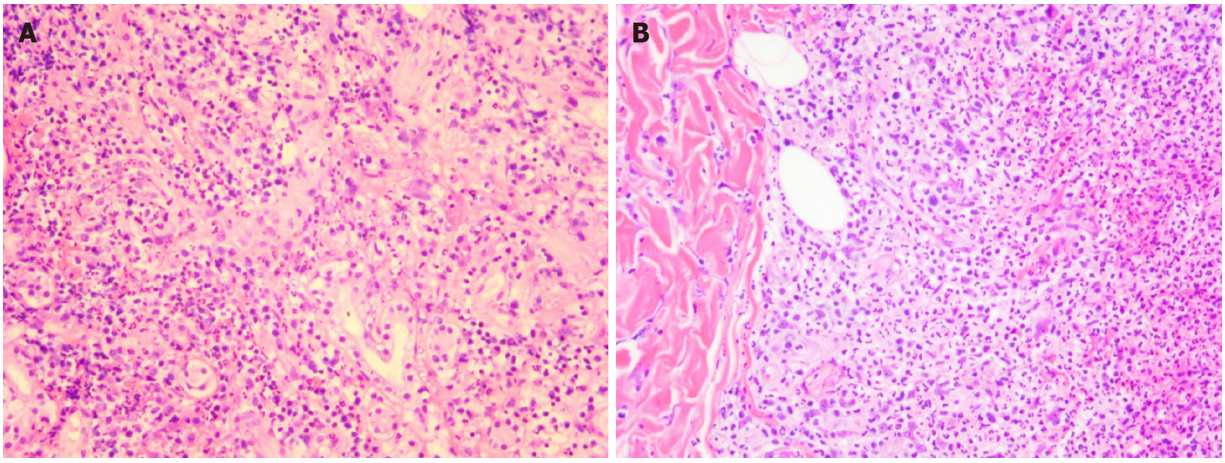

Figure 3 Skin and airway mucosa biopsy (magnification 100 ×). A: Skin mucosa biopsy; B: Airway mucosa biopsy. Subepidermal bullous formation containing scattered neutrophils and eosinophils.

pristinamycin, hydroxychloroquine, quinolones, sulfonamides, terbinafine, ketoconazole, diltiazem, and fluconazole[4-8]. Additional causes of AGEP, such as contact with mercury[9], have also been described. In this case, the patient took traditional Chinese medicine before the onset of the disease; thus, the possibility of AGEP caused by heavy metals in the traditional Chinese medicine or the traditional Chinese medicine itself should be considered.

AGEP is a self-limited disease with a short course and good prognosis. In severe cases, glucocorticoids should be administered. The mortality rate is less than 5%, and death is usually caused by multiple organ dysfunction and diffuse intravascular coagulation. Patients with a high risk of death are generally associated with other diseases or extensive skin lesions and mucous membrane involvement[10]. This patient had already experienced respiratory failure and airway mucosa involvement on admission; however the clinicians had an insufficient understanding of AGEP. A glucocorticoid was administered for more than 10 d after onset of the disease, and the overall prognosis was satisfactory.

CONCLUSION

AGEP is a self-limited disease with a good prognosis. This case represents a rare clinical feature of AGEP and an important finding for clinicians. Doctors should pay attention to possible AGEP, when a patient has rashes and fever.

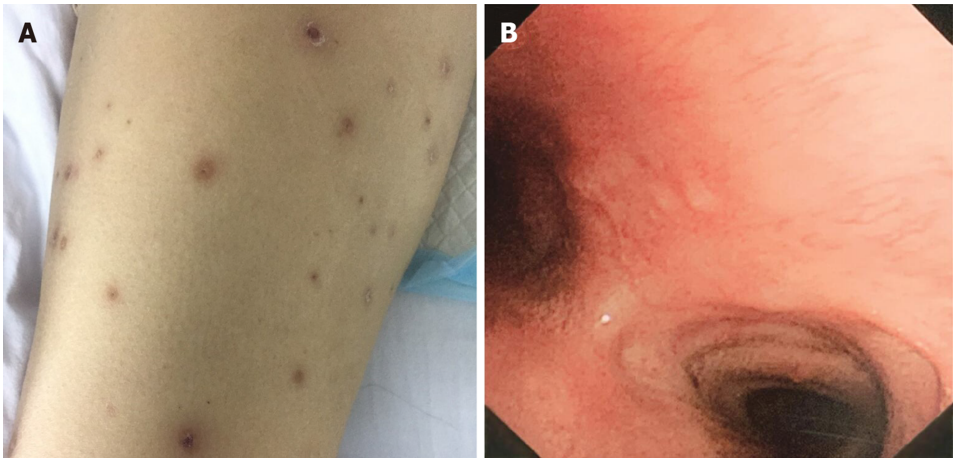

Figure 4 Leg rashes following transfer from the intensive care unit and final tracheoscopy. A: Leg rashes following transfer from the intensive care unit; B: Final tracheoscopy. Main airway was unobstructed.

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