# Exotropia in a pediatric patient with rhabdomyolysis caused by an insect sting

Won Jae Kim, Saeyoon Kim<sup>1</sup>, Myung Mi Kim

Various ocular and systemic reactions have been associated with insect sting. However, insect stings have been rarely reported to cause exotropia and diplopia. We encountered exotropia in a 6-year-old child with rhabdomyolysis of the left lower extremities caused by an insect sting. Exotropia and diplopia developed within 1 day after the sting and improved completely

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Departments of Ophthalmology and <sup>1</sup>Pediatrics, Yeungnam University College of Medicine, Daegu, South Korea

Correspondence to: Prof. Myung Mi Kim, Department of Ophthalmology, Yeungnam University College of Medicine, #317-1 Daemyung-Dong, Nam-Gu, Daegu 705-717, South Korea. E-mail: mmk@med.yu.ac.kr

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1 week after the onset of symptoms. Clinicians should be aware of the potential for the development of exotropia in patients with insect stings, which requires careful follow-up.

Key words: Exotropia, rhabdomyolysis, sting

Insect stings can result in local allergic and severe systemic reactions, including rhabdomyolysis, renal failure, liver injury, and coagulopathy.<sup>[1-4]</sup> A range of ocular reactions such as corneal edema, conjunctivitis, cataract, and optic neuritis have been reported after insect stings.<sup>[1]</sup> These reactions were attributed to direct trauma and potential toxic effects. However, insect sting has been rarely reported to cause strabismus. To the best of our knowledge, this paper describes the first reported pediatric case in a 6-year-old boy who developed exotropia with rhabdomyolysis caused by an insect sting.

## Case Report

Informed consent was obtained from patient's parent. A 6-year-old boy consulted our pediatric ophthalmology department for horizontal diplopia. He was admitted to the Pediatric Department and diagnosed with rhabdomyolysis after an insect sting. His mother reported that he had been stung

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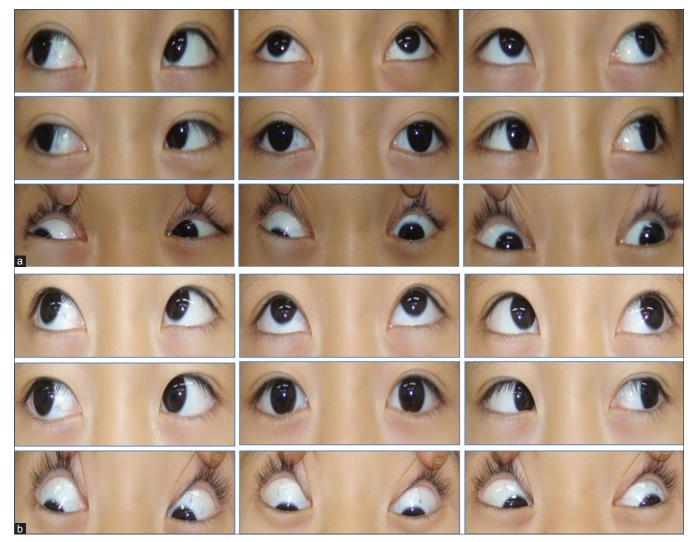
by an insect at a camping site 5 days earlier. Unfortunately, definitive identification of the offending insect was not possible.



**Figure 1:** (a) Diffuse edema and ecchymosis of the left lower extremity without sign of necrosis. (b) A single sting mark was visible on the left knee (arrow)

He reported feeling a mild itching sensation at the time at the sting site, the left knee. However, the patient had generalized edema, and ecchymosis extending from the sting site to the proximal and distal part of the left lower extremity on the evening that he visited the medical service [Fig. 1a and b]. One day later, he began complaining of horizontal diplopia. The initial laboratory results were as follows: White blood cell count of 13,260/µl (normal: 4000-10,000), blood urea nitrogen of 11.42 mg/dl (8-23), serum creatinine of 0.46 mg/dl (0.6-1.5), creatine phosphokinase (CPK) of 16,464 IU/L (57-374), lactate dehydrogenase of 1229 IU/L (150-550), myoglobin of 265 ng/ml (19-92), aspartate aminotransferase of 456 IU/l (10-35), and alanine aminotransferase of 173 IU/L (0-46). CPK level, which is a reliable and sensitive indicator of muscle injury, was significantly increased as shown by the laboratory findings.

No abnormal findings were observed in the blood coagulation studies, and the result of the urine analysis was normal. His mother reported that he had no prior history of strabismus or objective diplopia. There was no family history of strabismus. The corrected visual acuity was 20/20 in both eyes. The color vision test using Ishihara plate was normal.



**Figure 2:** (a) Images of the case in nine diagnostic position of gaze, demonstrating extropia of the right eye without definitive adduction limitation. (b) One week after its onset, the exotropia had improved and the patient had no diplopia

His pupils were equal, round, and showed normal reactions. He demonstrated 25 prism diopters (PDs) exotropia in the primary position without any limitation of adduction [Fig. 2a]. In the right and left gaze position, he had 25 PD exotropia. The Worth 4-dot test showed diplopia at a distance and near. No other abnormal ocular findings were noted. Cerebrospinal fluid analysis revealed normal findings, and cultures were negative. Brain magnetic resonance imaging (MRI) including three-dimensional constructive interference in the steady state (CISS) through the brainstem to visualize the cisternal segments of the cranial nerves and the orbit showed no abnormalities. One week after the onset of diplopia, he felt that the diplopia had improved. He was orthotropic under prism and alternate cover test and had no diplopia [Fig. 2b]. The worth 4-dot test showed fusion at a distance and near.

# Discussion

Several reports have shown that insect stings can cause both local and systemic reactions.<sup>[1-5]</sup> Among these reactions, strabismus is rare. Bee or tick stings have been reported to be associated with esotropia and abducens nerve palsy.<sup>[6,7]</sup> To the best our knowledge, there are no previous reports of exotropia in a child after an insect sting. In addition to strabismus, our patient showed rhabdomyolysis in his left lower extremity. Rhabdomyolysis after an insect sting in children can cause acute renal failure and other potentially fatal symptoms, necessitating immediate treatment, and close monitoring.<sup>[2,3]</sup> Fortunately, the patient showed stable improvement without further complications. In this case, the insect could not be identified. Since there have been reports of exotropia after snake bites, the possibility of snake bite was also considered initially.<sup>[8,9]</sup> However, the possibility of snake bite was ruled out for the following reasons. First, no fang mark suggesting a snake bite was observed. One bite mark was observed on the knee. Second, previous cases of exotropia after snake bite usually have a bite site at the finger rather than the knee as in the present patient. Third, the patient's mother had been with her child at the camping site and did not report seeing a snake. Tick or fire ants were the suspected offender in the current case, based on the sting mark and the parent's statements. The precise mechanism of strabismus onset after a sting is unclear. The possibility of an acute structural problem in orbit was ruled out through MRI with CISS sequences. The possibility of a potential toxin affecting the neuromuscular junction, as in the cases of strabismus after a snake bite, was considered. The extraocular muscles have a higher ratio of nerve fibers to the extraocular muscle fibers (1:6-1:12) compared to the large proximal limb muscles (1:2000).[8] Therefore, the extraocular muscle is particularly susceptible to neurotoxins. These extraocular muscle characteristics were also verified by extraocular muscle paresis after injection for botulinum toxin around the eye for cosmetic or medical use.<sup>[10]</sup> In this case, it is interesting that the patient showed exotropia without incomitance. The previous study reported comitant exotropia without obvious duction limitations after snakebite; the authors postulated that all extraocular muscles might have been equally affected by the toxin, which could lead to the appearance of a comitant motility deficit.<sup>[8]</sup> We agree with their hypothesis base on ocular motility findings. In conclusion, exotropia and diplopia can develop after an insect sting, and may gradually improve. Unusual reactions related to insect stings, including both ocular and systemic reactions, can occur. Clinicians need to be aware of the potential for the development of exotropia in patients with insect sting, which may require careful follow-up.

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#### **Conflicts of interest**

There are no conflicts of interest.

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