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Case Report

An Atypical Case of Lacrimal Sac Fistula Located on the Temporal Side of the Lateral Canthus

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We report a case of congenital lacrimal sac fistula located on the temporal side of the lateral canthus. A systemically healthy 1-year-old girl came to the outpatient clinic with a complaint of tearing on the temporal side of the right lateral canthus since birth. On examination, a small orifice was found in the skin on the temporal side of the lateral canthus. There was no evidence of inflammation or swelling within the opening. Surgeons carried out an operation under general anesthesia. They passed a probe through the lacrimal orifice and advanced it toward the lacrimal sac. Next, they introduced saline to the inferior punctum and found that it drained to the lateral fistula. The lower lid stretched as the dissected fistula was pulled. After the operation, the patient was free of the symptom. This paper is to report a case of congenital lacrimal sac fistula located on the temporal side of the lateral canthus.

Key Words: Lacrimal sac, Lateral, Fistula

A congenital lacrimal sac fistula is a lacrimal duct dysplasia in which epithelium from the lacrimal sac or the lacrimal duct is connected to the skin [1]. In most cases, the lesions are unilateral and located at the inferolateral to medial canthus. Most cases are asymptomatic and do not progress. Therefore most fistulas are incidentally discovered during routine clinical examinations [2]. The authors now report an atypical case of congenital lacrimal sac fistula located on the temporal side of the lateral canthus.

Case Report

A systemically healthy 1-year-old girl came to the outpatient clinic with a complaint of tearing on the temporal side of the right lateral canthus since birth (Fig. 1). Watering increased on weeping or blinking. On examination, a small orifice was found in the skin at the temporal side of the lateral canthus. There was no evidence of inflammation or swelling around the opening. An operation was carried out under general anesthesia. In the saline syringing test, saline drained through the lateral lacrimal sac fistula. We certified that the fistula was connected to the lacrimal drainage system by probing the orifice of the fistula and finding that the probe ran straight in the direction of the lacrimal sac (Fig. 2A). The skin was incised and a fistulectomy was performed 2 cm away from the orifice. The lower lid stretched as the dissected fistula was pulled (Fig. 2B and 2C). The remnant tract was closed with 6-0 vicryl and cauterization using a monopolar device. The skin was closed with 6-0 plain catgut. A histologic examination of the lacrimal fistula showed stratified squamous epithelium.

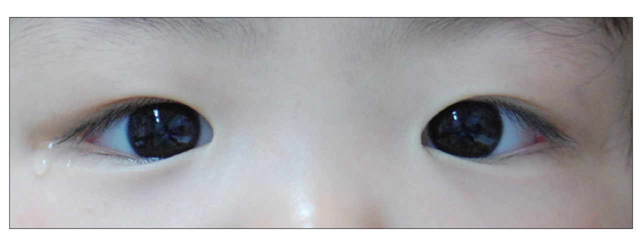

Fig. 1. A small orifice was found in the skin on the temporal side of the right lateral canthus and the watering increased on weeping or blinking.

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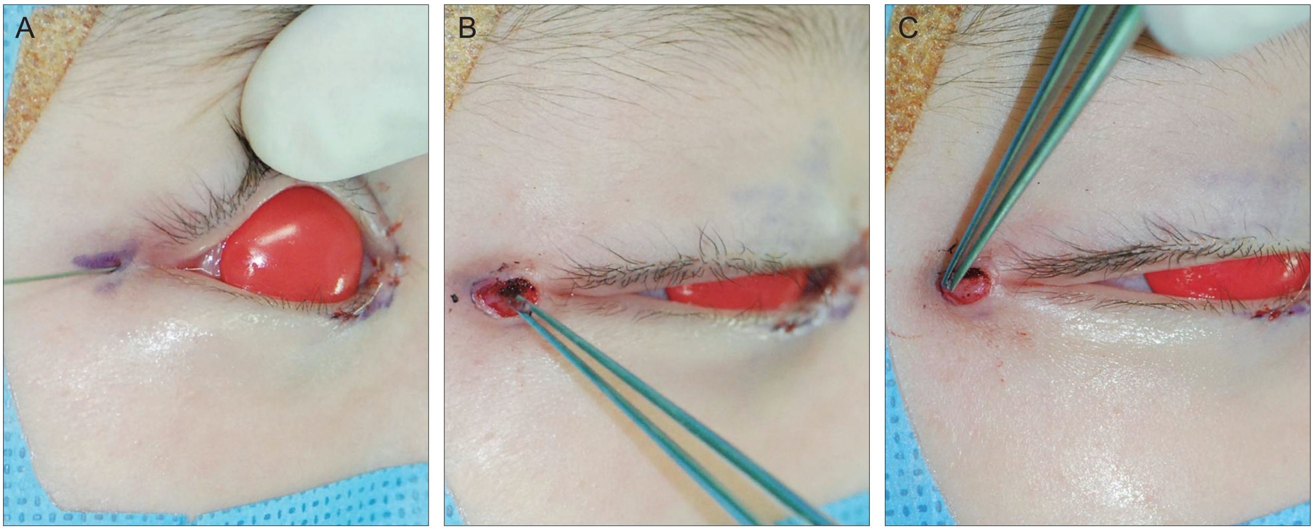

Fig. 2. (A) The probe ran straight in the direction of the lacrimal sac. (B) The fistula was exposed, and (C) the lower lid was stretched as the dissected fistula was pulled laterally.

There was no lacrimal gland tissue. After the operation, the patient was free of the symptom.

Discussion

The nasolacrimal apparatus arises embryologically from a cord of surface ectoderm that invaginates between the maxillary and frontonasal processes, giving rise to the canaliculi proximally, and the lacrimal sac and nasolacrimal duct distally. Canalization of the buried ectodermal cord occurs throughout the length of the nasolacrimal apparatus. Incomplete separation of the cord from the surface epithelium or abnormal out-budding of the buried ectodermal cord can result in supernumerary punctae and canaliculi [3].

Patients with congenital lacrimal sac fistula commonly present with an epiphora or discharge. In spite of the congenital nature of these fistulas, the clinical presentation is often delayed for many years after birth due to the evaporation of small amounts of discharge [4]. In this case, the patient had a history of epiphora since her birth, so the case was found early.

In the past, electrocauterization and a simple skin excision were usually performed to alleviate symptoms, but these methods have not been used recently because of their poor success rates. Jeong [5] had good results with fistulectomy and silicone tube intubation when there existed lacrimation through the eye. Song et al. [6] showed that if there is no lacrimation through the eye, a simple fistulectomy is good enough for a successful result. In this case, the patient had only lacrimation through the lacrimal fistula, and there was no combined nasolacrimal duct obstruction. Fistulectomy alone caused the patient to be free of symptoms, and she is currently on observation.

Most of the congenital lacrimal sac fistulas have been unilateral and located at the inferolateral to the medial canthus [4]. In this case, however, the patient had a unilateral fistula at the temporal region of the lateral canthus. A lateral lacrimal sac fistula can be confused with congenital lacrimal gland fistula. In this case, a dacryocystogram and other radiologic studies were not performed. The authors identified the lateral lacrimal sac fistula based on three observations: 1) saline drained through the lateral lacrimal sac fistula; 2) the probe ran straight to the sac through the lateral lacrimal sac fistula; and 3) the lower lid was stretched when the dissected fistula was pulled. Thus we report a case of a congenital lacrimal sac fistula located on the temporal side of the lateral canthus.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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