

Mirror Image of Unilateral Lacrimal Fistula in Monozygotic Twins: A Case Report and Review of The Literature

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Abstract

Background: To report a case of mirror unilateral lacrimal fistula in monozygotic twins and to review the literature.

Materials and Methods: Full ophthalmic examination including pupillary light reflex, Hirschberg test, ocular movements, face photographs was performed. Pediatrics consultation was requested.

Results: We report an identical twins, 6-months age - boys, with unilateral lacrimal fistula. For each twin, there was lacrimal fistula, located on the right for the first twin and on the left for the second one. The parents noted that the pits were present since birth. It was asymptomatic in each twin from the outside. There were no ocular and systemic associations with fistula.

Conclusion: We report first case to our knowledge of mirror unilateral lacrimal fistula in monozygotic twins.

Keywords: Lacrimal fistula; Lacrimal anomaly; Monozygotic twins; Mirror images in twins

Introduction

Hereditary lacrimal sac fistula is rare developmental condition. Fistulas of lacrimal sac are visible as small openings in the skin of medial canthus. Fistula connects common canaliculus, lacrimal sac or nasolacrimal duct to skin. In some cases, fistula ends blindly in the subcutaneous tissue. The symptoms are epiphora and mucopurulent reflux. Sometimes there may be no symptoms [1,2].

Several cases of mirror pathologies have been described, mainly concerning the renal and dental organs rarely ocular structures. The appearance of such mirror pathologies may be the consequence of the rise of a genetic or environmental factor during the acquisition of the zygote, occurring before its split into two monozygotic embryos [3].

We report a case of asymptomatic mirror unilateral lacrimal fistula in monozygotic twins with the review of the literature.

Materials and Methods

Full ophthalmic examination including pupillary light reflex, Hirschberg test, ocular movements, face photographs was performed. Pediatrics consultation was requested.

Results

Case report

We report an identical twins, 6-months age - boys, with unilateral lacrimal fistula. For each twin, there was a lacrimal fistula, located on the right for the first twin (Figure 1) and on the left for the second one (Figure 2). The parents noted that the pits had been present since birth. They had never complaint of tearing from the outside. The opening of fistula was seen to be obliterated. It was asymptomatic in each twin. Detailed systemic evaluation was performed by the pediatricist. There were no ocular and systemic associations with fistula. Visual acuity was fix-follow-maintain (FFM) positive in each eye of each twin. Cycloplegic refraction was +1.50 (+0.50α90) bilaterally in the first twin (right located fistula) and +1.75 (+0.25α90) bilaterally in the second twin (left located fistula). The pupils were equal in size with normal pupillary light reflex. Anterior segment and fundus examinations were

normal. Ocular examinations showed normal adnexal structures, eyes were orthophoric to Hirschberg testing, and eye movements were conjugate and normal.

Discussion

Studies of twins assume a special place in genetics because of the possibility of comparing genetic and environmental factors. Distinction



Figure 1: The presence of lacrimal fistula's pit at the right side.

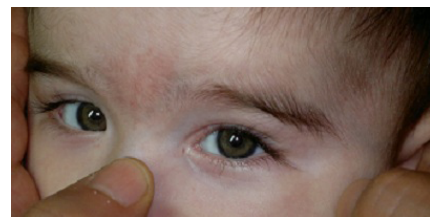


Figure 2: The presence of lacrimal fistula's pit at the left side.

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of twins as identical or fraternal is important in these studies. Identical twins account for 8% of all twins. The genome and sex are the same in monozygotic twins. The precise method of determining zygosity is standard genotyping. However, a mirror image, which is specific to monozygotic twins, is a strong sign on decision of identical or not. A mirror image is the same features, normal or anomalous, in opposite organs, which is estimated to occur in 25% of monozygotic twins [4]. Why some twins are mirror imaged and not other is still a mystery to scientist. No one knows why the egg splits and so no one knows why some twins mirror each other and others don't. There is not a lot of solid scientific evidence and study on mirror imaging so a lot is anecdotal. One explanation is that later fission of monozygotic twins makes particularly prone to this process [5]. The cleavage of a single fertilized ovum usually occurs between the 3rd and 8th days of gestation [6]. If this event is initiated after the 8th day, a mirror image may appear [7]. There are a few reports about mirror image unilateral eye anomalies in identical twins. These are generally about strabismus and refractive errors. Intermittent esotropia and myopic anisometropia [8], congenital esotropia and concordant hypermetropia [9], congenital esotropia and discordant refractive errors [10], myopic anisometropia [11], keratoconus [3], optic nerve dysplasia and anisometropia [12] are the mirror images of the unilateral eye disorders that detected in identical twins in different studies. We couldn't find any report on mirror lacrimal anomalies in monozygotic twins. So, this report may be first in the literature.

Congenital or hereditary lacrimal system anomalies are uncommon. There are a few reports on hereditary lacrimal fistula [13-15]. The hereditary cases commonly have been reported bilaterally unlike the congenital cases [15]. However, our cases have unilateral fistula. Hereditary fistula generally have an autosomal dominant inheritance [15]. Maden et al. suggested an autosomal recessive inheritance in their case series [16]. In a detailed history of our cases, there were no lacrimal fistulas in their parents (mother and father), third brother, grandfathers (2 persons), grandmothers (2 persons), uncle, aunts (2 persons) and cousins (5 persons). We examined their mother, father, third brother, uncle, 1 aunt and 3 cousins. The other relatives were not examined. We learned from the history that the other relatives didn't have lacrimal fistulas. In our report, we think that the findings suggest that the twins may have recessive inheritance in lacrimal fistula.

The hereditary lacrimal sac fistula may be associated with some ocular or systemic developmental anomalies. Systemic associations with lacrimal fistula are so rare and it required to be proven by genetic methods. Systemic disorders such as CHARGE syndrome (coloboma, heart anomalies, choanal atresia, retardation of growth and development, genital and ear anomalies) [17], Down syndrome [18,19], Waardenburg-Klein syndrome [20], VACTERL syndrome (vertebral anomalies, anal atresia, cardiac malformations, tracheo-esophageal fistula, renal and limb anomalies) [21], ectrodactyly-ectodermal dysplasia-clefting syndrome [22], hipospadias [23], preauricular fistula [24] have been seen with lacrimal fistula in previous studies. Ocular associations with lacrimal fistula such as double lower lid puncta [24], punctum agenesis [17,24,25] canalicular atresia [25], nasolacrimal duct stenosis [26], orbital meningocele [27], mucocele [28], strabismus [23], hypertelorism [23] have been reported before.

In our report, the pediatrics examination was normal in each twin. The ocular examination including adnexal structures, Hirschberg test, and eye movements was normal in each patient. We didn't feel the need to perform further investigations like imaging or genetic methods.

The management of lacrimal fistula includes observation,

cauterization, simple closed excision of fistula, excision with intubation and/or dacryocystorhinostomy. In the case of lacrimal fistula without symptoms, the observation is the best choice [29,30]. In our case, we decided to follow-up for a period of time because of no symptoms.

In conclusion, this case is the first report to our knowledge of mirror unilateral lacrimal fistula in monozygotic twins. We think it will be helpful as a data to further genetic investigations in the future.

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