An unusual twinning moment – A rare occurrence of unilateral congenital dacryocystocele in a pair of monozygotic identical twins

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A pair of 3-day-old twins were brought to the outpatient department for evaluation regarding swelling near the eye [Fig. 1a and b]. The swellings were tense in nature with no associated watering. An endoscopic nasal evaluation was obtained to rule out intranasal extension, and they were diagnosed with unilateral congenital dacryocystocele, which is very rarely reported in twins. Conservative management in the form of topical antibiotics and Crigler massage was initiated, following which there was complete resolution [Fig. 1c and d].

Congenital dacryocystocele arising in monozygotic (identical) twins are very uncommon and have been rarely reported in the literature.[1-3]

Discussion

The identical twins presented with similar dacryocystocele in a mirror image fashion (left side in twin 1 and right side in twin 2). The mirror image clinical presentation implies that anatomical factors involved in the development of the congenital dacryocystocele may have a genetic preponderance in the twins. From an anatomical point of view, the embryogenesis of a congenital dacryocystocele is due to prenatal developmental failure of the nasolacrimal duct (NLD) system to cannulate. Though the prenatal diagnosis of congenital dacryocystocele supports the argument for a developmental error, the role of a genetic basis for this anatomical malformation cannot be ruled out.

Barham et al. supported the genetic theory and concluded that congenital dacryocystocele has a genetic basis in their case series of a set of identical twins with bilateral dacryocystoceles.[3] Traquair highlighted a pattern of familial inheritance in 11% of patients in a large study involving the etiology of dacryocystitis.[4]

Further literature review revealed three cases of familial NLD obstruction. However, two of the three cases were associated with syndromes that resulted in NLD obstruction and were not directly related to isolated congenital duct obstruction. The first case involved a mother and daughter with branchio-oculo-facial syndrome, which resulted in congenital NLD obstruction.[5] The second case involved a brother and sister with lacrimal puncta agenesis, which resulted in congenital NLD obstruction.[6] The third case involved two non-twin female siblings with unilateral isolated right dacryocystoceles. This study concluded that the likely cause for NLD obstruction in the non-twin siblings was a sporadic embryologic event.[7]

The basis for an argument for a genetic basis and not familial basis in our case series lies in the lesions occurring in a set of monozygotic twins rather than non-twin siblings. One possible explanation for why a genetic predisposition has not been better elucidated is that, there is significant underreporting of this finding. The significant underreporting may be due to a large percentage of spontaneous resolution of this condition.[7][11] Another school of thought as to why genetic predisposition has not yet been established says that, multiple members of a familial group with subclinical findings would never be evaluated or studied.

Though there have been few reported cases suggestive of familial predisposition, in this manuscript, we have reported a set of monozygotic twins with congenital dacryocystocele, which further strengthens the argument for a genetic basis of this entity.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Figure 1: (a and b) Photographs showing a bluish-gray cyst below the medial canthus on the left side (OS) of twin 1 and right side (OD) of twin 2, respectively. (c and d) Follow-up photographs at 4 months showing complete resolution in twin 1 and twin 2, respectively.

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Conflicts of interest
There are no conflicts of interest.

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