Infantile Perineal Protrusion in Two Monochorionic Twins

Paola Cavicchioli, MD1  Enrico Valerio, MD2  Mario Cutrone, MD1

1 Department of Pediatrics, Ospedale Dell’Angelo, Mestre, Venice, Italy
2 Department of Woman and Child Health, Medical School, University of Padova, Padova, Italy


Address for correspondence  Enrico Valerio, MD, Department of Woman and Child Health, Medical School, University of Padova, Via Giustiniani, 3 35128 Padova, Italy
(e-mail: enrico.valerio.md@gmail.com; enrico.va@inwind.it).

Case Report

Two female monochorionic-monoamniotic twins were born from cesarean section at 37+2 weeks. Pregnancy was regular, and fetal echographies have been always normal. Maternal serologies were negative. At first clinical evaluation 3 hours after birth the same pinkish, rubbery, stem-like perianal lesion was noted in both the twins (►Fig. 1), consistent with infantile perineal protrusion (IPP). First-born twin (on the left in the picture) presented with IPP on the left border of the anal orifice, while second-born twin (on the right in the picture) presented with median-line IPP anterior to the anus.

A literature review numbers approximately 100 reports of IPP.1,2 This condition has been classically classified into three categories: congenital/familiar (i.e., female sex, positive parental history of IPP), acquired (mainly due to constipation), and associated with lichen sclerosus et atrophicus.2

Conclusions and Final Remarks

This case report describes, for the first time, the presence of IPP in monochorionic-monoamniotic twins, supporting the existence of hereditary/genetic factors in the developing of this condition.

Received April 21, 2014  Accepted after revision May 27, 2014  Published online August 11, 2014

ISSN 2157-6998.

Copyright © 2014 by Thieme Medical Publishers, Inc., 333 Seventh Avenue, New York, NY 10001, USA.
Tel: +1(212) 584-4662.

Keywords
► infantile perineal protrusion
► monochorionic twins
► congenital disease
► neonatology

Abstract

Case Report  Two female monochorionic-monoamniotic twins showed the same kind of infantile perineal protrusion (IPP) at birth. Lesions in both twins progressively healed until resolution in 6 weeks' time; none of the twins have manifested, till date, alvus disturbances.

Discussion and Literature Review  A literature review numbers approximately 100 reports of IPP. This condition has been classically classified into three categories: congenital/familiar (i.e., female sex, positive parental history of IPP), acquired (mainly due to constipation), and associated with lichen sclerosus et atrophicus.

Conclusions and Final Remarks  This case report describes, for the first time, the presence of IPP in monochorionic-monoamniotic twins, supporting the existence of hereditary/genetic factors in the developing of this condition.
Differential diagnoses of IPP encompass sexual abuse, perianal Crohn disease, hemorrhoid disease, rectal prolapse, and hemangiomas.\(^1,3,5,9\)

Constitutional IPP most often resolves in a few weeks’ time from initial diagnosis, thus warranting a conservative approach\(^3\); less frequently, this form of IPP may persist over several years.\(^2,4\)

Acquired IPP may regress upon effective treatment of constipation\(^6,7\) or, more rarely, may persist despite alvus normalization.\(^2\)

LSA-related IPP treatment may encompass topical steroid therapy,\(^10\) although spontaneous resolution of the IPP has been documented also in this condition.\(^8\)

Consistently with constitutional IPP, lesions in both presented twins progressively healed until resolution in 6 weeks’ time; none of the twins have manifested, to date, alvus disturbances.

This report is of educational value since it contains the first observation of congenital IPP in two monochorionic-monoamniotic female twins at birth: such evidence supports the existence of hereditary/genetic factors in the development of the IPP.

Conflict of Interest
None.

References
9 Tsunemi Y, Matsushita T, Takahashi T, Tamaki T. Childhood capillary hemangioma presenting as infantile perianal protrusion. Dermatology 2003;207(4):408–409