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Case Report

Dissimilar Effects of Thalidomide in Dizygotic Twins

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Among 93 cases of thalidomide embryopathy ascertained in Brazil, one pair of female dizygotic twins was found. One of them presented complete paraxial hemimelia in the right arm while the other showed a triphalangeal thumb at the left hand and had duodenal stenosis. Of the eight dizygotic twin pairs exposed to thalidomide in utero described thus far, only four were concordant for the type of malformation.

Key words: Thalidomide, Embryopathy, Teratogenesis, Twins

INTRODUCTION

Not much is known about the effects of thalidomide in twins. As far as we know, only one pair has been adequately described [2]. These twins were dizygotic (DZ), male and female, and their mother had taken the drug during the first four months of pregnancy. The boy had bilateral scapular phocomelia and a midline hemangioma on the forehead, while the girl had only anomalies of the thumb, but she was born with duodenal atresia and rectoperineal fistula. Pfeiffer and Kosenow [3] mentioned that, out of six pairs of DZ twins they had examined, four had the same type of malformation, but they gave no details. A report is hereby made of the occurrence of another pair of differentially affected twins ascertained during a general survey of Brazilian thalidomide victims.

We have had the privilege of gaining access to the records of the Brazilian Association of Thalidomide Victims, founded with the main objective of promoting legal actions to obtain indemnification for damages from the pharmaceutical industry and the Brazilian government. Of a total of 204 cases, originating from 10 Brazilian States, we selected 93 cases almost certainly affected by the thalidomide embryopathy. Two of them are the twins whose description will be presented below. Detailed medical data are available on these records and additional information was obtained by mail from the twins' father, who is himself a physician.

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Fig. 1. M.J.C., the twin who has complete paraxial hemimelia in her right arm.

CASE REPORT

Two twin girls, M.J.C. and M.C.C., were born with congenital malformations on 28 August, 1962, at Belo Horizonte, the capital of the Brazilian State of Minas Gerais. The mother had taken a sedative drug called Slip, whose basis was thalidomide, manufactured by Lafi Laboratory, during the first three months of pregnancy. The twins are the daughters of nonconsanguineous parents and have a sister and a brother, born respectively one year and three years after them, both normal. The father reports other cases of twins among his ancestors, but denies other instances of congenital malformations in his or his wife's families. He also informs that, independently of the different lesions, the twins had never been confounded by close relatives or strangers. M.J.C. shows right arm radius and thumb aplasia, ulna hypoplasia, and clubhand. Besides this complete paraxial hemimelia, she presents congenital dislocation of the shoulder, agenesis and hypoplasia of arm and forearm muscles, and elbow damage (Fig. 1).

The only limb malformation found in her co-twin (M.C.C.) is a triphalangeal thumb at the left hand; but she was born with duodenal stenosis due to an annular pancreas (Figs. 2, 3). M.J.C. was examined by Dr. E. Marquardt, from the Orthopedic Clinic of the University of Heidelberg in 1971, who confirmed the diagnosis of thalidomide embryopathy.

COMMENTS

In any attempt to explain the diversity of type or intensity in the malformations found in a pair of twins exposed to a given drug in utero, it should be kept in mind that the twins'

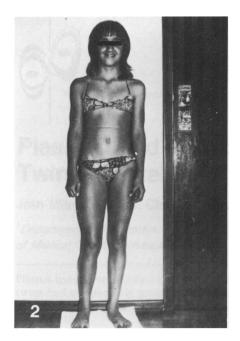




Fig. 2. M.C.C., the other twin; her triphalangeal thumb has already been surgically corrected. Note the scar above the umbilicus, consequence of the surgery made due to the duodenal stenosis.

Fig. 3. X ray of M.C.C.'s left hand obtained before the surgery. Note the triphalangeal thumb.

development is not entirely synchronous. Besides that, different blood supplies and microenvironmental differences can contribute to the variability of effects. But, according to Lenz [1], the hypothesis that human embryos are genetically differentially predisposed to the teratogenic action of thalidomide cannot be completely excluded. Considering the pair described here and the seven others reported by other authors, we would have concordance in only half of the DZ pairs of twins simultaneously exposed to thalidomide in utero.

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