LETTERS TO THE EDITOR

Drug induced VATER association: is dibenzipin a possible cause?

The VATER complex is a non-random combination of three or more vertebral, anorectal, tracheo-oesophageal, radial, and renal anomalies in a neonate, with abnormalities of the heart or kidneys. The VATER association has been associated as a possible cause1 and, recently, lead intoxication2 and lovastatin administration3 have also been implicated. We describe the first case of VACTERL association in a neonate whose mother had been treated with dibenzipin.

The proband is the second child of a 25 year old woman who suffered from depression and had been treated with dibenzipin (Victoril) (80 mg x 3/day) throughout her pregnancy. The mother did not smoke or drink alcohol or coffee, and had not suffered from any infection during pregnancy. The parents are not related, and the father is healthy. The family history was unremarkable. The mother had not taken any medication during her first pregnancy and gave birth to a boy with no abnormalities. During the second pregnancy (1992), ultrasound and z fetoprotein measurements were not performed. The fetal heart rate near term showed a normal rhythm.

The 40 week term infant, born in February 1993, with a weight appropriate for gestational age (3260 g), presented with oesophageal atresia,1 lead1 and, recently, lumbosacral hemivertebrae, dextroptosis of the heart, and right cryptorchidism. Brain and kidney ultrasound were normal. Karyotyping and G banding studies were normal. The oesophageal malformations were surgically corrected by end to end anastomosis and fistulectomy. At the age of 3 months, physical and neurological development was normal.

Dibenzipin is a tricyclic antidepressant, rarely used during the first trimester. Because of the very limited data on dibenzipin usage in pregnancy, a precise risk estimation cannot be performed.

It is noteworthy that neither extensive epidemiological studies in humans nor experimental data have shown evidence of an association between the use of tricyclic antidepressants in pregnancy and birth defects,1 and the teratogenic effect of these drugs remains uncertain and contradictory. The VATER complex is one of the more common patterns of multiple malformations in newborns.2 Prenatal exposure to exogenous sex hormones,1 lead1 and, recently, lovastatin1 might have a teratogenic effect. Although it is impossible to establish a causal relationship between prenatal dibenzipin exposure and VATER on the basis of a single case report, the present report adds another possible teratogenic agent and raises the possibility of a drug induced type of VATER complex. A large prospective multicentre collaborative study is needed to clarify this issue further.

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References

Kyphomelic dysplasia

A six year follow up of a child with kyphomelic dysplasia has shown some interesting and previously undescribed features. This case was originally described by Temple et al.1

Kyphomelic dysplasia is a rare condition with a generalised abnormality of skeletal growth. It is caused by rhizomelic and mesomelic limb shortening with short, broad, and bowed long bones, the femora being most severely affected. There is metaphyseal flaring and irregularity in infancy, causing prominent joints with restricted mobility. The ribs are short and flared. Variable features include micrognathia, midface hypoplasia, long philtrum, facial haemangioma, abnormal rib number, mild platyspondyly, increased acetabular angles, and skin dimples over bony prominences.

To date 11 cases of kyphomelic dysplasia have been reported.

The subject, a male child, remained mildly dysmorphic at the age of 3 years 3 months. Rhizomelic shortening of the limbs with anterior bowing of the thighs was still evident, but had improved since infancy. His gait was abnormal with a tendency to toe walk on the left and he began to complain of intermittent pain in the knees.

Velocardiofacial syndrome and DiGeorge sequence

I was interested to read the series of articles published in the Journal of Medical Genetics1-3 regarding phenotypic overlap of the velocardiofacial syndrome and DiGeorge sequence, preceded by Judith Hall's Editorial advocating the use of the acronym CATCH 22, as suggested by Wilson et al.4 The attention to velocardiofacial syndrome and 22q11 is very exciting and the authors are to be congratulated on this collective body of material.

I would like to respond to a number of points which, in my opinion, point out the important role of clinical diagnosis, even in conditions where molecular analysis has yielded positive results. The articles of Greenberg,2 Goldmuntz et al,5 Driscoll et al,4...
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