Acrocyanosis due to imipramine

Transient neonatal hyperthyroidism and maternal thyroid stimulating immunoglobulins

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SUMMARY Serum thyroid stimulating hormone binding inhibitor immunoglobulins (TBII) and thyroid stimulating antibody (TSAb) concentrations were measured in three pregnant women with hyperthyroidism and then in their infants. The results suggested that TBII concentrations in infants in the neonatal period or from mothers during the third trimester of pregnancy showed a good correlation with the development of neonatal hyperthyroidism.

It is now widely believed that the transient thyroid overactivity in infants born to mothers with hyperthyroidism is a result of the placental transfer of thyroid stimulating immunoglobulins from the mother to the infant. The immunoglobulins in the sera from some patients with Graves’ disease were shown, by the radioreceptor assay system, to inhibit the binding of thyroid stimulating hormone to its receptor sites and were designated as thyroid stimulating hormone binding inhibitor immunoglobulins (TBII). It was also shown that immunoglobulins in the patient’s sera stimulated the production of cyclic AMP in human thyroid tissues and were known as thyroid stimulating antibodies (TSAb).

We studied three unrelated infants who were born to women with hyperthyroidism, and one of them developed typical transient neonatal hyperthyroidism. We measured serum TBII and TSAb concentrations in these infants and their mothers.

Case reports

Family 1 The mother developed hyperthyroidism at 26 years old when she was in her 32nd week of pregnancy. From the 34th week until delivery she had been maintained euthyroid with methimazole. A girl (3330 g) was born at 39 weeks’ gestation. On the 8th day of life she was noted to be hyperactive and had developed diarrhoea. Tachycardia was noticed on the 10th day when her body weight was 3100 g. Thyroid studies on day 8 showed that she had hyperthyroidism; she had serum concentrations of triiodothyronine, thyroxine, free thyroxine, and

References


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thyroid stimulating hormone of 8.6 nmol/l, 527.6
nmol/l, 350.8 pmol/l, and <1.0 mU/l, respectively.
Treatment with propylthiouracyl and Lugol’s solu-
tion was started on day 13. Her condition and
thyroid function improved gradually and the treat-
ments were stopped on day 59. Her physical and
mental development and the thyroid function tests
gave normal results at 10 months of age.

Family 2 The mother was first diagnosed as having
hyperthyroidism at 29 years old. Two years after the
diagnosis she received partial thyroidectomy be-
cause of a poor response to antithyroid drugs. After
the surgery she had been maintained euthyroid with
thyroxine supplement. A girl was born at 39 weeks’
gestation. Her development and thyroid function
were normal during 10 months of follow up.

Family 3 The mother developed hyperthyroidism at 22
years old when she was four months pregnant.
Treatment was started with methimazole but she
remained hyperthyroid during the rest of her preg-
nancy. A boy (2590 g) was born at 39 weeks’
gestation and his thyroid function, detected in the
cord blood, was normal. Thyroid hormone concen-
trations gradually decreased until 2 weeks of age
when serum concentrations of triiodothyronine,
thyroxine, and free thyroxine were 1.3 nmol/l,
47.7 nmol/l, and 8.0 pmol/l, respectively. The hormone
concentrations returned to the normal ranges at 1.5
months of age without treatment. Throughout this
period thyroxine binding globulin concentrations
were normal and thyroid stimulating hormone
concentrations were always less than 1.0 mU/l. At
4 months of age his development and thyroid func-
tions were normal.

Methods

Serum samples from the three pregnant women and
their infants were stored at -20°C until assayed.

During follow up some of the specimens taken at
certain intervals from the same infant were com-
bined for TBII and TSAb assays.

Serum TBII was assayed by the method of
Shewring and Smith3 using a commercial kit (Japan
Travenol, Japan). Serum TSAb concentrations were
assayed by the method of Kasagi et al.4

Results

In the infant in family 1 who developed neonatal
hyperthyroidism, both TBII and TSAb concentra-
tions were high at birth (table 1) and gradually
decreased by day 50 (table 2). The concentrations of
TBII and TSAb in the mother of this infant at the
third trimester of pregnancy were close to the
concentrations in the neonate.

The mother in family 2 had detectable TBII and
TSAb concentrations in the first trimester of preg-
nancy and the infant had detectable TSAb but
undetectable TBII concentrations in the combined
sera obtained on day 4 and 11 (table 1). No TSAb
was detected in the infant after the neonatal period
(table 2).

The mother in family 3 had detectable TBII and
slightly high TSAb concentrations in the second
trimester and detectable but further decreased
concentration of TBII in the third trimester of

Table 2 Concentrations of TBII and TSAb in the infants of family 1 and family 2 after birth

<table>
<thead>
<tr>
<th>Age (days)</th>
<th>Family 1</th>
<th></th>
<th>Family 2</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>0</td>
<td>13/17*</td>
<td>27/36*</td>
<td>43</td>
</tr>
<tr>
<td>TBII</td>
<td>58.7</td>
<td>45.7</td>
<td>28.0</td>
<td>25.1</td>
</tr>
<tr>
<td>TSAb</td>
<td>5.3</td>
<td>3.4</td>
<td>2.1</td>
<td>1.8</td>
</tr>
</tbody>
</table>

Control values: TBII, <10%; TSAb, <1.0 mU/l.
*Serum samples obtained on two different days were combined for assays.

Table 1 Concentrations of TBII and TSAb in the mothers during pregnancy and their infants at birth (family 1 and 3) or
neonatal period (family 2)

<table>
<thead>
<tr>
<th>Family</th>
<th>Mothers</th>
<th>Neionates</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Trimester 1st</td>
<td>2nd</td>
</tr>
<tr>
<td></td>
<td>TBII</td>
<td>TSAb</td>
</tr>
<tr>
<td>1</td>
<td>*</td>
<td>*</td>
</tr>
<tr>
<td>2</td>
<td>33.4</td>
<td>22.0</td>
</tr>
<tr>
<td>3</td>
<td>*</td>
<td>*</td>
</tr>
</tbody>
</table>

*Assay not done.

Control values: TBII: <10%; TSAb: <1.0 mU/l.
Transient neonatal hyperthyroidism

Severe combined immunodeficiency syndrome, tissue transplant, leukaemia, and Q fever

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SUMMARY A child born with severe combined immunodeficiency, who was immunoreconstituted by a fetal liver and thymus transplant, developed acute lymphoblastic leukaemia in the donor cell line. During remission she contracted acute Q fever, which gave rise to unexpected complications. Early treatment of the Q fever might have altered the subsequent events and prevented her death.

Q fever is a sporadic infection and is an occupational hazard of men working with domestic sheep and cattle. The clinical features of the acute and chronic form in adults are well described. Reports in children are rare with less than 50 notifications from 1975–81. Heard et al reported five cases in immunocompromised adults. We describe a girl, who was 6 years old at the time of her death, who had been immunoreconstituted by transplantation with fetal liver and thymus at 9 months old because of severe combined immunodeficiency (SCID). Acute lymphoblastic leukaemia (ALL) subsequently developed in the donor cell line. During treatment she developed acute Q fever from which she ultimately died. The advantage of co-

References


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