Gynaecomastia is a source of significant psychological and emotional distress in adolescent males. Currently, the main mode of treatment is mastectomy, while literature on pharmaceutical management is limited. Tamoxifen is a selective estrogen receptor modulator which has shown considerable success in treatment of pubertal gynaecomastia. However its efficacy in management of gynaecomastia due to endocrine causes is yet unknown.

This case series evaluates clinical outcome of 3 adolescent males with PAIS having gynaecomastia who were treated with tamoxifen.

Tamoxifen appears to slow down progression of breast development and reduce psychological distress in patients with moderate gynaecomastia up to 3 years, allowing deferment of irreversible procedures like mastectomy until the child is old enough to take part in decision making process.

Use of tamoxifen at the onset of gynaecomastia appears to give better results in controlling the breast growth.

### Table 1. Pre-treatment and post-treatment clinical features of the patients

<table>
<thead>
<tr>
<th>Age of onset of gynaecomastia</th>
<th>Patient 1</th>
<th>Patient 2</th>
<th>Patient 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at starting tamoxifen</td>
<td>12 months</td>
<td>12 months</td>
<td>14 years</td>
</tr>
<tr>
<td>Current age</td>
<td>16 years</td>
<td>16 years</td>
<td>16 years</td>
</tr>
<tr>
<td>Pre-treatment</td>
<td>16 years</td>
<td>18 years</td>
<td>12 years</td>
</tr>
<tr>
<td>Post-treatment</td>
<td>16 years</td>
<td>18 years</td>
<td>12 years</td>
</tr>
</tbody>
</table>

1. Clearly a source of significant psychological and emotional distress in adolescent males.
2. The main mode of treatment is mastectomy, while literature on pharmaceutical management is limited.
3. Tamoxifen is a selective estrogen receptor modulator which has shown considerable success in treatment of pubertal gynaecomastia. However its efficacy in management of gynaecomastia due to endocrine causes is yet unknown.

**Results**

**Case 1**
- 16-year-old boy with genetically confirmed PAIS, presented with bilateral gynaecomastia at 11 years of age.
- Pre-treatment palpable glandular breast tissue measured 8x10cm bilaterally (Tanner stage III).
- Tamoxifen 10mg daily was started at the age of 12 years 8 months and continued for 4 years without side effects.
- Tamoxifen therapy curtailed further progression of breast development, reduced granularity and maintained breast development in Tanner stage III during the first 3 years of treatment.
- Reduction in psychological distress in the patient and the parents was also reported.
- However during last 6 months of treatment, rapid growth of bilateral breasts was noted (stage IV).
- Mastectomy is currently being considered due to persistent gynaecomastia (Figure 1) and patient wishes.

**Cases 2 and 3**
- Two brothers with 46XY DSD (presumed PAIS), currently aged 14 and 16 years, born with micropenis and penoscrotal hypospadias, were started on tamoxifen.
- Elder sibling (case 2) presented with bilateral gynaecomastia at 14 years 2 months of age (5x5 cm bilaterally, Tanner stage III).
- Tamoxifen 10mg daily was started and continued for 2 years. Breast tissue reduced to 3x3 cm after 1 year and remained stable thereafter (Figure 2a).
- Younger brother (case 3) presented with bilateral gynaecomastia (right breast 3 x 4 cm; left breast 2.2 cm) at 13 years (Figure 2b.) and was commenced on tamoxifen 10 mg daily.
- Gradual regression of glandular breast tissue (right breast 2.5 x 3 cm, left breast non-palpable) was noted after 1 year of treatment.
- Both brothers report a reduction in psychological distress, lack of adverse effects and would like to continue therapy for longer.

**Conclusions**

Tamoxifen appears to slow down progression of breast development and reduce psychological distress in patients with moderate gynaecomastia up to 3 years, allowing deferment of irreversible procedures like mastectomy until the child is old enough to take part in decision making process.

Use of tamoxifen at the onset of gynaecomastia appears to give better results in controlling the breast growth.

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