(Case series) Use of tamoxifen in the management of gynaecomastia in three adolescent boys with 46 XY DSD.

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INTRODUCTION

- Gynaecomastia is a source of significant psychological and emotional distress in adolescent males[1].
- Currently, the main mode of treatment is mastectomy, while literature on pharmaceutical management is limited[2].
- 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.[2]

AIM

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

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 16 and 14 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 4cm; left breast 2x2 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.

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

	Patient 1	Patient 2	Patient 3
Age of onset of gynaecomastia	11 years	14 years	13 years
Age at starting tamoxifen	12 years 8 months	14 years 2 months	13 years
Current age	16 years 9 months	16 years 3 months	14 years 4 months
Pre-treatment Right side (Tanner stage) Left Side (Tanner stage)	III (8x10cm) III (8x10cm)	III (5x5cm) III (5x5cm)	II (3x4 cm) II (2x2 cm)
Tamoxifen dose (mg/d)	10	10	10
Duration of treatment (months)	49	25	16
Post-treatment Right side(Tanner stage) Left Side (Tanner stage)	IV IV	II (3x3cm) II (3x3cm)	II (2.5x3cm) I (Non palpable)

Figure 1. B/L gynaecomastia in patient 1 at 16 years 7 months (after the rapid growth in the last 6 months of treatment)

Figure 2a.

Figure 2b.

Figure 2a. B/L gynaecomastia of patient 2 at 15 years (after 1 year of treatment)
Figure 2b. B/L gynaecomastia of patient 3 at 13 years (pre-treatment).

CONCLUSIONS

- Tamoxifen appears to slow down progression of breast development and reduce psychological distress in patients with moderate gynaecomastia up to 3 years, allowing deferement 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.

REFERENCES

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