Original Study

Improving Self-Esteem in Women Diagnosed with Turner Syndrome: Results of a Pilot Intervention

Paul M. Chadwick DClinPsy, MA [1], Arlene Smyth [2], Lih-Mei Liao PhD, MSc [3,*]

[1] *Clinical and Health Psychologist, Royal Free Hampstead NHS Trust, London, UK*
[2] *TSSS, Clydebank, Scotland, UK*
[3] *University College London Hospitals, London, UK*

A B S T R A C T

Study Objective: To evaluate a brief intervention to improve the self esteem of women diagnosed with Turner syndrome (TS).
Design: Prospective observational study.
Setting: Turner Syndrome Support Society, UK.
Participants: 30 women aged 18-60 years.
Intervention: A 1-day psychology workshop targeting problems of self-esteem in women diagnosed with TS. The workshop drew on cognitive-behavioral therapy and narrative therapy skills and emphasized increased self-awareness of interpersonal difficulties and improved capacity for self-management.
Main Outcome Measures: Rosenberg Self-esteem Scale (RSS); Hospital Anxiety and Depression Scale (HADS); bespoke user experiences questionnaire.
Results: All 30 women provided baseline data, 27/30 provided immediate post-intervention data and 22/30 provided follow-up data at 3 months. The intervention improved RSS and HADS scores at 3 months.
Conclusion: Generic skills-based psychological interventions have the potential to be adapted to provide brief and low-cost interventions to improve self-esteem and reduce psychological distress in women diagnosed with TS.
Key Words: Turner syndrome, Disorders of sex development, Self-Esteem, Psychology

Introduction

Turner Syndrome (TS) affects 1 in 2,500 live female births[1] and is the second most common chromosomal condition after Down syndrome. It is caused by the total or partial deletion of 1 of the X chromosomes. Mosaicism is relatively common; in rare cases, some Y material may also be present.[2] TS affects the functions of a number of body systems. It is associated noticeably with primary amenorrhea, infertility, and shorter than typical stature. There are other variable effects including, for some individuals, subtle impairment in hearing[3] and non-verbal learning.[4] In the longer term, there are elevated risks of developing certain cardiovascular and metabolic diseases.[5]

The medical consequences of TS vary widely between individuals and the focus of medical management changes across the lifespan. In childhood and adolescence, clinical management is focused upon maximizing adult height and future bone health, as well as pubertal development and reproductive potential. In adulthood, the focus is more on direct management of resultant conditions such as infertility, cardiovascular disease (congenital malformations, aortic root dilation, and hypertension) and osteoporosis, among others. The monitoring of audiological functioning and cardiovascular risk is important throughout the life span.[5]

TS was recently included in a new classification scheme developed by a consensus group who put forward the term disorders of sex development (DSD) to include all "congenital conditions in which development of chromosomal, gonadal, or anatomic sex is atypical.[6]" Among other changes, the consensus statement emphasized multi-disciplinary team work and the integration of expert psychological care "to promote positive adaptation." A subsequent widely endorsed UK care standard document[7] further emphasized integral psychological care in multi-disciplinary management of initial presentation of DSD in infancy and adolescence. However, the formulation of psychological interventions and accountability have been left to the imagination of individual practitioners.[8]

Research in the psychological wellbeing of TS-affected women is scarce, although problems of negative self-evaluation have been identified.[9] Fewer women diagnosed with TS engage in intimate relationships compared to women of comparable age, but the reasons for this have not been ascertained. Evidence-based psychological interventions for these problems exist,[10] but documentation of their implementation as part of quality care for TS and other DSDs is extremely rare. The emphasis on the psychological challenges of TS is paralleled by the conspicuous absence of the development of effective remedial interventions.

The current pilot study was the result of dialogues over the years about these concerns between 2 specialist clinical psychologists and a UK national user group (TSSS UK). In this article, we describe the process and outcome of a pilot study to explore the effects of a single episode day-

The authors indicate no conflicts of interest.

* *Address correspondence to:* Lih-Mei Liao, PhD, University College London Hospitals, London, WC1E 6DB, UK. Phone: +448451555000 ext 72511
E-mail address: lih-mei.liao@uclh.nhs.uk (L.-M. Liao).

1083-3188/$ - see front matter
http://dx.doi.org/10.1016/j.jpag.2013.09.004

long group psychological intervention for women affected by TS.

Materials and Methods

Development of the Intervention

The goal of the psychological intervention, that is, to improve self-esteem was decided on collaboratively between all of the authors. The eventual intervention was designed by the 2 senior authors (P.M.C., L.M.L), both clinical psychologists and took the form of a full-day workshop. The workshop emphasized self-management skills (as opposed to 'talking therapy') and drew on 2 prominent therapeutic approaches in clinical psychology. Cognitive-behavioral therapy is a 'directive' therapeutic approach that encourages skills development in identifying and modifying unhelpful patterns of thoughts, emotions and action.[11] Narrative therapy encourages individuals to identify self-diminishing self stories and formulate alternative self views.[12]

The workshop was delivered for a total number of 30 participants. During the workshop, a number of individual and group exercises enabled the participants to identify ways in which TS might have affected their self-evaluation. The participants were then directed to practice with each other a number of techniques to challenge negative self-perceptions and to contemplate taking realistic steps to overcome avoidance.

Participants

The 30 workshop participants were recruited through a mail out distributed to members of the UK TSSS. The inclusion criteria were: (1) a diagnosis of TS; (2) aged 18 or above; (3) able to take part in the group independently of partners, family members, carers, or friends without TS. The mean age of the participants was 34.5 years (SD = 14.1) ranging from 18 to 60 years. The majority (29/30) self-identified as White UK. Seven (23.3%) were married, 5/30 (16.7%) were not married but in a stable relationship, and 18/30 (60%) were currently not in a relationship. The women currently in a relationship reported having been with their current partner for an average of 8.14 years.

Outcome Measures

This was a prospective evaluation whereby participants completed questionnaires before and at the end of the workshop and again 3 months later. The questionnaires, comprising the following measures, were mailed out with a stamped addressed envelope 3 months following the completion of the workshop. All participants consented to anonymized dissemination of the work.

Rosenberg Self-esteem Scale (RSS)[13]

This 10-item scale is a well-established measure of self-esteem in adult women. Participants are asked to indicate the degree to which they agree with a series of statements. Each item is scored 1 or 0 thus the maximum score is 10 with higher scores representing higher self-esteem.

Table 1
Frequencies of Answers to Questions about the Workshop

Questions About the Workshop	Responses			
	Strongly Agree	Agree	Disagree	Strongly Disagree
I found the workshop interesting	17	6	-	-
I found the workshop helpful	12	9	2	-
I am able to view TS in a more positive way since attending the workshop	5	16	1	1
Attending to workshop has helped me to be more open about TS	1	16	5	1
Attending the workshop has helped me to feel less different to other women	6	13	4	-
Attending the workshop has helped me to feel that difference can be a positive thing	7	14	2	-

Hospital Anxiety and Depression Scale (HADS)[14]

This 12-item scale was designed to assess levels of anxiety and depression in individuals with a physical illness. The HADS has 2 subscales: Anxiety and Depression. Participants are asked to check 1 of a series of responses which best reflects their experience of symptom over the past month. Higher scores represent a greater level of symptomatology. The scale also yields a threshold above which a score is considered clinically significant.

User Feedback at 3 Months

The 3-month follow-up questionnaire also included a user experiences questionnaire (written for this purpose; see Table 1 for the items) that sought participants' views of the workshop. In particular they were asked to rate the helpfulness of the workshop on a scale of 1 (Not At All Helpful) to 10 (Extremely Helpful), and indicate the extent to agreement or disagreement with a series of statements regarding the workshop and its subsequent effects on a scale of 1 (Completely Disagree) to 10 (Completely Agree). They were also invited to offer any comment about their experiences of the intervention.

Results

Statistical Analyses

The distribution of scores for the principal measures (RSS, HADS Anxiety, HADS Depression) were tested for normality at all time points using the Shapiro-Wilk test. All of the data were normally distributed thereby justifying the use of t-tests to assess the significance of changes in self-esteem (pre- and immediately post-intervention, pre-intervention, and 3-month follow-up) and HADS Anxiety and Depression (pre-intervention and 3-month follow-up) for matched pairs of individuals.

The 3-month follow-up data were not available for 8/30 participants. Compared to those who provided follow-up data (N = 22), the 8 nonrespondents had lower pre-intervention Anxiety (4.3 vs 9.1) and Depression (3.6