

## Empirical Article

**Cost-effectiveness analysis of a psychosocial web-based intervention for adolescents distressed by a visible difference: Results from a randomized controlled trial in Norway**JOHANNA KLING,<sup>1</sup>  LARS ASPHAUG<sup>2</sup> and KRISTIN B. FERAGEN<sup>1</sup><sup>1</sup>Centre for Rare Disorders, Oslo University Hospital, Oslo, Norway<sup>2</sup>Clinical Trials Unit, Oslo University Hospital, Oslo, Norway

Kling, J., Asphaug, L. & Feragen, K. B. (2022). Cost-effectiveness analysis of a psychosocial web-based intervention for adolescents distressed by a visible difference: Results from a randomized controlled trial in Norway. *Scandinavian Journal of Psychology*.

Adolescents with a visible difference to the face or body (e.g., due to a congenital condition, illness, or injury), may be at risk of elevated psychological distress. Young Person's Face IT (YPF), a web-based psychosocial intervention, has displayed effectiveness, but no previous study has specifically evaluated its cost-effectiveness. The aim of our study was to investigate whether YPF could be a cost-effective alternative for psychosocial support to adolescents distressed by a visible difference, relative to care-as-usual (CAU). Within the context of a randomized controlled trial in Norway, 102 participants (43% boys) were allocated to intervention ( $n = 55$ ) or waiting list control group ( $n = 47$ ). Mean age was 13.9 years ( $SD = 1.71$ ; range 11–18), and all self-identified as experiencing distress related to a visible difference. Participants answered questionnaires including measures of health-related quality of life and social anxiety at baseline and 3-month follow-up. A health economic evaluation using the method of cost-utility analysis was performed, including quality-adjusted life-years (QALYs). Results indicated that the incremental cost-effectiveness of YPF was 63,641 Norwegian kroner per QALYs gained, which is well within the acceptability threshold in the Norwegian healthcare sector. Hence, YPF could potentially be considered a cost-effective intervention for adolescents experiencing distress related to a visible difference, but more research is needed that includes comparisons of YPF to other health- and societal resources and long-term follow-ups. Our study also constitutes an addition to research as, compared to interventions for somatic diseases, there is a lack of studies exploring the cost-effectiveness of psychological interventions.

**Key words:** Cost-effectiveness analysis, internet intervention, quality-adjusted life-years, randomized controlled trial, adolescents, visible difference.

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## INTRODUCTION

Self-perceptions about one's appearance is a common source of social and psychological distress, especially during adolescence (Ricciardelli & Yager, 2016), and particularly for those involuntarily having an appearance that noticeably deviates from the norm (Crerand, Rumsey, Kazak, Clarke, Rausch & Sarwer, 2020). Young Person's Face IT (YP Face IT; YPF; Williamson, Hamlet, White *et al.*, 2016), a web-based self-guided psychosocial intervention, has demonstrated promising results in reducing psychological distress among adolescents with a visible difference (Kling, Zelihić, Williamson & Feragen, 2022; Zelihić, van Dalen, Kling *et al.*, 2022). However, health economic evaluations of YPF and similar interventions for this population are lacking.

*Visible difference: definition, prevalence, and consequences during adolescence*

A range of conditions, both congenital and acquired, may affect facial or bodily appearances and lead to what is commonly referred to as a visible difference (Rumsey & Harcourt, 2007). Congenital conditions include skin conditions (such as psoriasis, haemangiomas or vitiligo) and craniofacial conditions (such as cleft lip and palate and syndromes, e.g., Treacher Collins). Acquired conditions may result from accidents, burns or medical interventions (such as cancer treatment). Due to the wide definition of visible difference, and the heterogeneity of the many

appearance-altering conditions, there are no certain prevalence rates for people living with a visible difference. However, previous estimates from the UK (Changing Faces, 2010) indicate that around 2.3% of the population may have a visible difference. Based on these estimates, as national statistics currently indicate there are approximately 400,000 adolescents aged 12–17 in Norway, around 9,000 of them may live with a visible difference (Statistics Norway, 2021).

Previous research suggest that adolescents with a visible difference may be at risk of elevated psychological distress, including anxiety (De Vere Hunt, Howard & McPherson, 2020; van Dalen, Dierckx, Pasmans *et al.*, 2020), and dissatisfaction with appearance (Huang & Su, 2021; King, 2018; Ngaage & Agius, 2018; Provini, Omandac, Bahrani, Aghdasi & Cordero, 2021). Moreover, adolescents with a visible difference who experience appearance concerns may also struggle with relational difficulties, such as fear of negative evaluations from others and increased concerns in peer and romantic relationships (Feragen, Stock, Sharratt & Kvaalem, 2016; Griffiths & Rumsey, 2012; Shapiro, Waljee, Ranganathan, Buchman & Warschausky, 2015). In addition, adolescents with a visible difference may experience stigmatizing or intrusive behaviors such as teasing, bullying, staring, or unwanted attention from others. Such negative social experiences have in turn been associated with reduced health-related quality of life and psychological adjustment (Masnari, Landolt, Roessler *et al.*, 2012; Masnari, Schiestl, Rössler *et al.*, 2013; Tiemens, Nicholas &

Forrest, 2013). Left untreated, this distress can potentially be aggravated and affect the adolescent's long-term well-being, which may lead to an increased economic burden for the healthcare system. For instance, in terms of anxiety disorders in the general population, if left untreated, they tend to cumulate depressive symptoms, school/work absence, distressing physical symptoms, substance abuse, and economic problems (Arikian & Gorman, 2001). Specifically related to appearance-related distress, if not addressed, social anxiety and dissatisfaction with appearance can lead to anxiety, depression, eating disorders, avoidance or over-indulgence in exercise, and reduced participation in society (e.g., Kanayama, Barry, Hudson & Pope, 2006; Stice & Shaw, 2003). Appearance concerns are also known to impact on healthcare decision-making and could influence choices regarding aesthetic and reconstructive surgery in adulthood, an economic burden from both social and health perspectives (Rumsey & Harcourt, 2004; Stice, 2002; Williamson & Rumsey, 2017). Consequently, early and effective interventions aimed at adolescents with a visible difference has the potential to reduce distress for the individual, and at the same time lower the overall costs to the healthcare system and society (Arikian & Gorman, 2001; Lépine, 2002).

#### *Interventions for adolescents distressed by a visible difference*

Support for adolescents with a visible difference consists of biomedical and psychosocial interventions. Studies have shown that biomedical interventions, such as medical and surgical procedures to correct or ameliorate appearance differences, may improve social confidence (Myhre, Råbu & Feragen, 2021). However, biomedical interventions do not guarantee enhanced psychosocial functioning and therefore psychological interventions have evolved as an adjunct or alternative to biomedical approaches (Bemmels, Biesecker, Schmidt, Krokosky, Guidotti & Sutton, 2013; Paraskeva, Tollow, Clarke *et al.*, 2021; Rumsey & Harcourt, 2007). Psychosocial support for adolescents with a visible difference typically involves a wide range of therapeutic approaches and techniques drawn from cognitive behavioral therapy (CBT), social skills training (SST), and acceptance and commitment therapy (Harcourt, Hamlet, Feragen *et al.*, 2018). Psychosocial interventions based on CBT and SST have specifically shown potential in improving psychosocial well-being in adolescents with a visible difference (Jenkinson, Williamson, Byron-Daniel & Moss, 2015; Williamson, Hamlet, White *et al.*, 2019), for example, adolescents with burn injuries (Blakeney, Thomas, Holzer, Rose, Berniger & Meyer, 2005), and children and adolescents with craniofacial and scarring conditions (Maddern, Cadogan & Emerson, 2006).

Regarding internet-based support, emerging research indicates that internet-delivered interventions (e.g., iCBT) generally can be effective in treating a wide range of psychological problems, (Barak, Hen, Boniel-Nissim & Shapira, 2008; Vigerland, Lenhard, Bonnert *et al.*, 2016), including conditions such as anxiety disorders (Stjerneklar, Hougaard, McLellan & Thastum, 2019), and depression (Topooco, Byléhn, Nysäter *et al.*, 2019), in samples of young people. Internet-delivered support also offers several benefits specific to adolescents experiencing challenges related to a visible difference. Clinical evidence suggests that

access to specialized psychosocial support and treatment is limited for those struggling with a visible difference (Harcourt *et al.*, 2018). Raising appearance issues face-to-face with healthcare professionals has also been shown to be experienced as sensitive and difficult (Gee, Maskell, Newcombe, Kimble & Williamson, 2019; Williamson, Harcourt, Halliwell, Frith & Wallace, 2010). Therefore, internet-based interventions may fill a gap in current healthcare provision by offering easily accessible support with greater anonymity and confidentiality (Griffiths & Rumsey, 2012).

#### *YP face IT: effectiveness and cost-effectiveness*

To date, YPF is the only web-based intervention developed for adolescents with a visible difference. The feasibility and acceptability of YPF has been explored in several countries (Feragen, 2017; Riobueno-Naylor, Williamson, Kogosov *et al.*, 2019; van Dalen, Pasmans, Aenderkerk *et al.*, 2021; Williamson *et al.*, 2019), indicating YPF as a promising intervention acceptable to adolescents. In addition, the effectiveness of YPF in improving body esteem and reducing symptoms of social anxiety, perceived stigmatization, and life disengagement, was also recently evaluated in a randomized controlled trial (RCT) with participants from the Netherlands and Norway (Zelihic *et al.*, 2022). The RCT showed that adolescents' in the intervention group had significantly lower levels of social anxiety post-intervention compared with the control group (with a medium effect size). Regarding clinically significant and reliable change, previous results in the Norwegian project (Kling *et al.*, 2022) showed that approximately 10% of all participating adolescents displayed a clinically significant and reliable improvement in social anxiety and/or body esteem following YPF. However, among participants with more time spent on the program and higher levels of distress at baseline, improvement was significantly higher. Hence, based on the few previous evaluations of YPF (Kling *et al.*, 2022; van Dalen *et al.*, 2021; Williamson *et al.*, 2019; Zelihic *et al.*, 2022), the intervention has displayed potential effectiveness. Much less is known about YPF's potential cost-effectiveness.

Increasingly, cost-effectiveness analysis has become an important addition to clinical outcome assessment in the evaluation of psychosocial treatments (Lombard, Haddock, Talcott & Reynes, 1998). Despite this, no previous study has specifically evaluated the cost-effectiveness of YPF. This is a common problem and there is a general knowledge gap with regard to the cost-effectiveness of intervention/prevention for mental health problems in adolescents in most European countries (Kilian, Losert, Park, McDaid & Knapp, 2010). However, the UK YPF pilot study (Williamson *et al.*, 2019) did include a feasibility assessment of the cost-effectiveness of YPF. Although limited by a small sample size and low completion rates in the resource use data collection, the authors concluded that YPF may prove to be cost-effective in the UK (Williamson *et al.*, 2019). Interestingly, Williamson *et al.* (2019) also noted that participants reported use of resources beyond the health and social care payer perspective, for example, with high costs for private counseling and cosmetic surgeries. With relevance for the present study, the UK YPF pilot study also concluded that it would be feasible to perform a cost-

effectiveness analysis within the framework of a future larger YPF RCT study (Williamson *et al.*, 2019).

### Aim

Health economic evaluation is an important part of overall psychosocial intervention evaluations, especially when the objective is to eventually implement the intervention into the healthcare system (Kilian, Losert, Park, McDaid & Knapp, 2010; Lombard, Haddock, Talcott & Reynes, 1998). The aim of the present study was to explore whether the web-based YPF intervention could be a cost-effective alternative for psychosocial support to adolescents distressed by a visible difference. Specifically, we aimed to evaluate the cost-effectiveness of YPF relative to care-as-usual (CAU), and within the context of a RCT in Norway.

## METHODS

The present study was conducted as part of a project exploring the effectiveness of the Norwegian version of YP Face IT (Trial registration number: NCT03165331; see Zelihić *et al.*, 2022). The study was conducted at the Centre for Rare Disorders, Oslo University Hospital, reviewed by the Regional Committee for Medical Research Ethics (Health Region South-East, reference number: 2015/2440), and accepted by the hospital's Data Protection Office.

### Participants and procedure

The present study included 102 participants (43% boys), randomized to intervention ( $n = 55$ ) or waiting list control group ( $n = 47$ , see Fig. 1). Mean age was 13.9 years ( $SD = 1.71$ ; range 11–18). Conditions resulting in a visible difference that were represented in the sample were: a craniofacial condition (64%), visible difference relating to body form (22%), skin condition (10%), and scarring (4%).

Recruitment took place between April 2019 and February 2021, and participants were recruited nationwide via specialist treatment units, local healthcare services, patient organizations, and through different media and social media platforms (Kling, Nordgreen, Kvaalem, Williamson & Feragen, 2021). All participants were screened for eligibility and the following inclusion criteria were used: (1) approximately 12–17 years with a visible difference leading to appearance-related distress and/or teasing/bullying; (2) access to the internet and a home computer/tablet; (3) reading level corresponding to that of a 12 year-old or higher; and (4) normal or corrected-to-normal vision. Exclusion criteria were: (1) a diagnosis of psychosis, eating disorder, clinical depression, and/or post-traumatic stress disorder, or within 12 months of traumatic injury; (2) learning disabilities that would hinder understanding of the program's content; and (3) currently receiving a psychological face-to-face intervention. The screening was performed during phone calls with potential participants and their primary care-givers, and thus inclusion/exclusion is based on self- and parental-reports. After screening, informed consent was obtained from all eligible participants. For participants <16 years, informed consent was also obtained from their primary caregivers. Subsequently, participants completed the baseline questionnaire and were randomized to either the intervention or the waiting list control group (Fig. 1).

### Intervention (YPF) and CAU

The YPF intervention was developed at the Centre for Appearance Research based at the University of the West of England, Bristol, UK, and the therapeutic content is based on CBT and SST. The content was developed in close collaboration with adolescents with visible differences, their parents, specialized clinical experts, and other health professionals (Williamson *et al.*, 2016, 2019). The program was translated into Norwegian in 2015 and

pilot-tested with adolescents in Norway (Feragen, 2017). The program is completed independently and provides support on how to adjust to common challenges related to having a visible difference and encourage adolescents to practice strategies through activities (for a more detailed description of the intervention, see Williamson *et al.*, 2016). YPF consists of seven weekly sessions plus an additional booster session, and each session is intended to take around 30–40 min (Williamson *et al.*, 2016). In our RCT, the intervention group received access to YPF immediately after randomization and answered a 3-month follow-up questionnaire after access to the intervention. On average, the participants in the intervention group completed 58.2% ( $SD = 37.3$ , range = 0–90) of the program. Number of sessions completed ranged from 0 to 8, with 85.2% of the intervention group participants completing at least one session, and a majority (53.7%) completing all eight sessions. After the 3-month follow-up, the control group also received access to YPF. Although this resulted in the study not being able to collect longer-term control data, it was considered necessary due to ethical reasons and in order to facilitate recruitment. For instance, the Norwegian pilot study showed that participants and parents were negatively disposed to participate without being given access to the program within a manageable future (Feragen, 2017).

Throughout the study, all participants received CAU. However, as there are no standardized psychosocial or psychological interventions for adolescents with a visible difference in Norway, CAU varies according to needs, resources, and expertise within local healthcare services, and includes, for example, routine consultations at the hospital for medical treatment. However, there is a general lack of psychosocial support for adolescents with a visible difference (Harcourt *et al.*, 2018), and for the vast majority of our participants, CAU was equal to no care related to appearance-related or social distress.

### Measures

**Social anxiety.** The total score of the social anxiety scale for adolescents (SAS-A; La Greca & Lopez, 1998) was used to assess social anxiety at baseline and follow-up. SAS-A contains 18 items (plus four filler items not included in the scoring), rated on a five-point scale ranging from 1 (*not at all*) to 5 (*all the time*). Items include “I worry about being teased” and “It's hard for me to ask others to do things with me.” The total sum of SAS-A has possible range from 18 to 90, where higher scores indicate higher levels of social anxiety. In the present study, participants' change scores from baseline to follow-up were dichotomized into “improvement” or “no improvement” based clinical cut-off values reported in the manual (La Greca, 1999), and methods for calculating clinically significant change reported by Jacobson and Truax (1991). SAS-A was translated from English to Norwegian for the purpose of the current project, using back-translation procedures (Brislin, 1970). In our study, overall internal consistencies (Cronbach's alphas) were  $\alpha = 0.95$  both at baseline and at follow-up (intervention group baseline:  $\alpha = 0.95$ ; intervention group follow-up:  $\alpha = 0.96$ ; control group baseline:  $\alpha = 0.94$ ; intervention group follow-up:  $\alpha = 0.95$ ).

**Health-related quality of life.** Health-related quality of life was measured by the Norwegian version 5-level EuroQol-5D (EQ-5D-5L) questionnaire (Herdman, Gudex, Lloyd *et al.*, 2011), a standardized instrument to measure generic health status for clinical and economic appraisal. The EQ-5D-5L has been validated in diverse populations in multiple countries, including clinical and non-clinical adolescent samples (e.g., Cheung, Wong, Samartzis *et al.*, 2016; Welie, Stolk, Mukuria *et al.*, 2022). EQ-5D-5L comprises five dimensions: mobility, self-care, usual activities, pain/discomfort and anxiety/depression. Each dimension is rated on one of the following five levels: no problems (e.g., “I have no pain or discomfort”), slight problems, moderate problems, severe problems and extreme problems (e.g., “I have extreme pain or discomfort”).

### Cost-effectiveness analysis

The Norwegian Directorate of Health (2012) advice that health economic evaluations should employ the method of cost-utility analysis. This type of

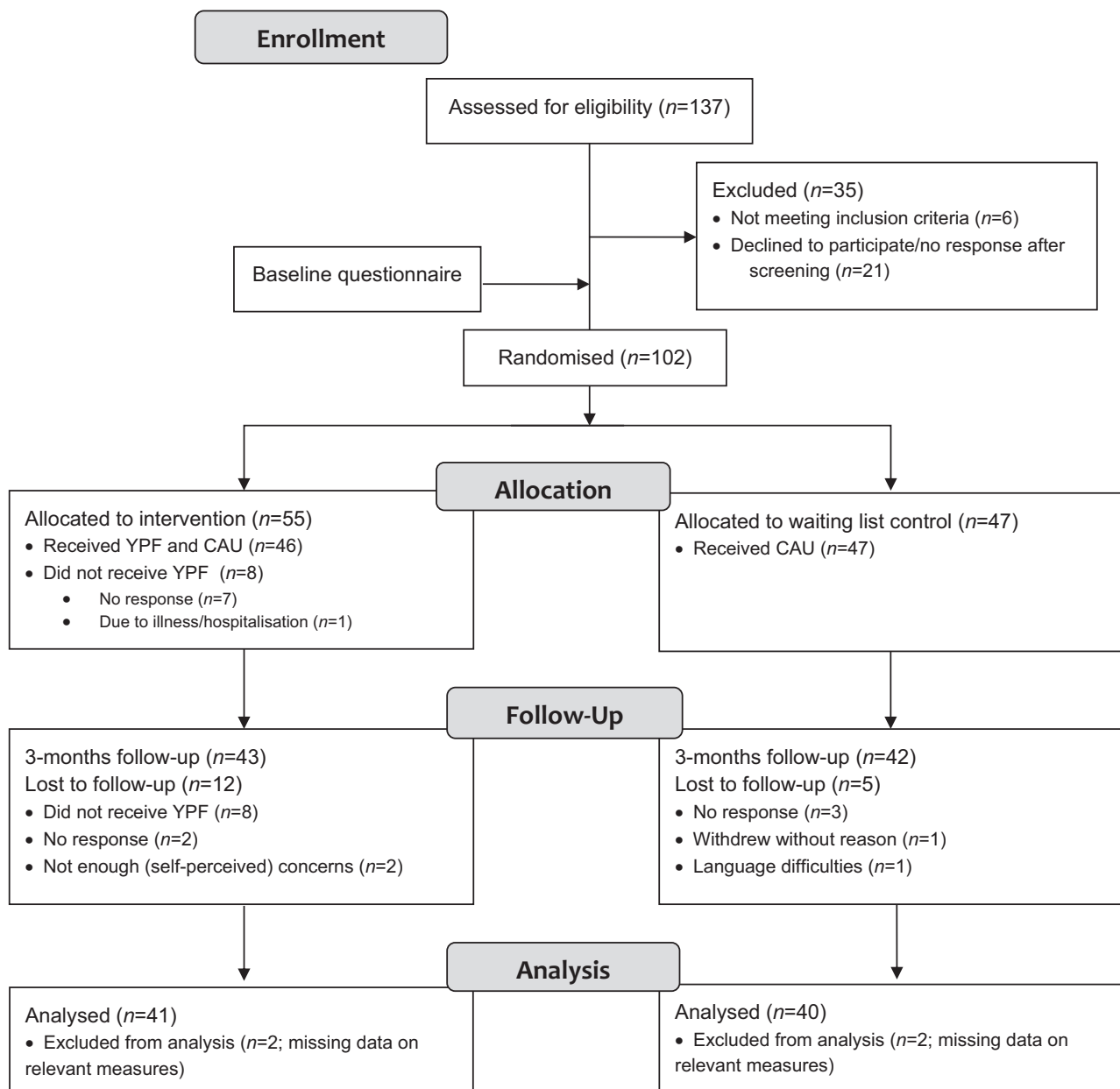


Fig. 1. Flow diagram based on CONSORT guidelines (Moher, Hopewell, Schulz *et al.*, 2010).

cost-effectiveness analysis uses quality-adjusted life-years (QALYs) as the measure of effectiveness. The aim of an economic evaluation is first to identify whether a new program is more effective, that is, whether it confers a QALY-gain, relative to the current treatment strategy. Further, as the healthcare sector has limited resources, an abundance of alternative uses of those resources, the incremental cost of introducing the new program must not exceed the cost-effectiveness acceptability threshold level. The outcome of interest for economic evaluations is the incremental cost-effectiveness ratio (ICER); the ratio of incremental costs to incremental QALYs gained. If the cost-per-QALY of the ICER is less than the acceptability threshold, the new program is considered a cost-effective alternative to the current strategy. This criterion can be summarized in the net-monetary benefit static:

$$\text{NMB} = W \times \Delta U - \Delta C > 0,$$

where  $W$  is the acceptability threshold value, and  $\Delta U$  is the incremental utilities gained and  $\Delta C$  is the incremental cost. If the

NMB is positive, the new program is considered to have acceptable cost-effectiveness for the decision maker, otherwise not.

*Quality-adjusted life-years.* QALYs denote the length of life weighted by the utility derived from living with a certain quality of life. In economics, utility is an individual's usefulness or enjoyment derived from the consumption of a good. We may measure an individual's utility loss from disability or disease (or utility gain from treatment) by their willingness to take risk to restore health (as in the standard gamble approach), or willingness to trade off time with disability from a shorter life in full health (as in the time trade off approach). As an alternative to deriving utility estimates for health states from the patients directly, an indirect approach is typically used in health technology appraisals in Norway. In the indirect approach, patients are asked to describe their health using generic questionnaires such as the EQ-5D. Following that, members of the general population are asked to trade off an amount of time in full health they consider equal to living a certain period in a health

state characterized by the patients' descriptions on the EQ-5D-5L. The respondents' average remaining time in full health relative to the whole period in the described health state is as such interpreted as the utility the described state. As there is currently no Norwegian general population valuation study for the EQ-5D-5L, we followed recommendations from the Norwegian Directorate of Health and used the results from the most recent British valuation study. Hence, we used British results for the EQ-5D-3L, mapped to the 5L questionnaire as per the recommendations of the British Decision Support Unit (National Institute for Health and Care Excellence; NICE, 2022). We implemented the mapping in R using the work of Hernández Alava, Pudney and Wailoo (2020).

To calculate QALYs we followed the 'area-under-the-curve' (AUC) approach (Manca, Hawkins & Sculpher, 2005). QALYs are calculated for each subject,  $i$ , by weighting their utility level at two time points  $u_i^0$  and  $u_i^1$  by the time between baseline and follow-up ( $t^1 - t^0$ ):

$$AUC_i = \left( \frac{u_i^0 + u_i^1}{2} \right) \cdot \frac{(t^1 - t^0)}{T}.$$

Dividing the sum of the utility levels by two assumes that the subject changes utility level halfway through the observation period. As  $t^0$  here is 0, and our time unit is in months, each subjects' utility was weighted by  $t^1 = 3$  and divided by  $T = 12$ , which aligns the outcome to the QALY timescale of 1 year.

Comparison of naive group averages at baseline revealed a difference prompting the need to adjust for difference in pre-treatment utility. We used multiple regression with QALYs as the dependent variable, and treatment-assignment and baseline utilities as independent variables. We tested several model specifications and selected a generalized linear model with Gaussian distribution and log-link (see Supplementary Appendix for regression diagnostics and comparisons of fit between other model specifications).

**Costs and YPF use.** Importantly, although we initially collected resource use data directly from our participants with the aim to compare resource use between the intervention group and the control group, these data are not included in the present study due to very low completion rates and poor data quality. Our estimated costs and usage of YPF are primarily based on experiences with YPF in the UK where the intervention has been implemented in health care services and through patient organizations (Williamson *et al.*, 2019; P. Tollow, personal communication, 14 March 2022). Hence, as the UK is the only country so far that has implemented YPF, we decided to base assumptions of service levels on their data in combination with data from national statistics in Norway, and recruitment data from the Norwegian RCT study. Operating costs for webhosting include cost of service, updates, and support (£365 = ~4,000 Norwegian kroner [NOK]/year). In addition, salary (including overheads and employer costs) for a limited part-time (<5% of full time employment) healthcare professional/assistant at Oslo University Hospital responsible for information and support to healthcare services and patient organizations providing YPF to their patients/members (~6,000 NOK/year, based on current salaries; Statistics Norway, 2022). The estimated number of users/year ( $n = 40$ ) is based on a combination of program use in the UK (with approx. Three users/month; P. Tollow, personal communication, 14 March 2022) and recruiting experiences in the Norwegian RCT study (with a mean recruitment of approx. Five participants/month; Kling, Nordgreen, Kvale, Williamson & Feragen, 2021). Since the program already exists in Norwegian, the translation/adaptation cost is not included in the calculations.

**Cost-effectiveness acceptability threshold.** There is no official cost-effectiveness acceptability threshold in Norway. The 2015–2016 Norwegian government white paper on priority setting in the healthcare sector discusses a base threshold of 275,000 NOK (Norwegian Government, 2016). The base threshold may increase up to 825,000 NOK, depending on the patient groups' expected loss of QALYs under the current treatment strategy relative to the age-matched general population (absolute prognosis loss). We followed the Norwegian Medicines

Agency's (2021) suggestion to calculate the absolute prognosis loss to the patients extrapolating with the control group's baseline QALY-level. The threshold-level corresponding to this prognosis loss according to the whitepaper's discussion was used as a proxy for the actual acceptability threshold.

**Decision uncertainty.** We used bootstrapping to assess the degree of uncertainty around the likelihood of cost-effectiveness (Glick, Doshi, Sonnad & Polsky, 2014). This allows assessing uncertainty without having to impose parametric assumptions on our highly skewed data. We performed 10,000 bootstrap samples from a multivariate normal distribution using the regression models' coefficients and its variance-covariance matrix. For each bootstrapped sample replica, we calculated the net monetary benefit using the assumed acceptability threshold. The likelihood of cost-effectiveness was calculated as the proportion of all bootstrapped samples in which the YPF conferred a positive net monetary benefit. We show this likelihood graphically for a range of acceptability threshold values in Fig. 2.

Data analysis was done using R, with post-analysis plotting using the 'dampac' package, and with code available in the Supplementary Appendix. As rates of missing data were very low, participants with missing data ( $n = 4$ ) were excluded and complete cases analyses were carried out (Fig. 1).

## RESULTS

### Improvement

The intervention group and the control group did not differ significantly in terms of baseline social anxiety ( $t [100] = -1.11$ ;  $p = 0.27$ ). In the intervention group, 13 participants (32%) had a clinically relevant decrease in social anxiety from pre- to post-intervention (i.e., improvement), and 28 (68%) did not display a clinically relevant decrease (i.e., no improvement). In the control group, five participants (12%) had a clinically relevant decrease in social anxiety from pre- to post-intervention (i.e., improvement), and 35 (88%) did not display a clinically relevant decrease (i.e., no improvement).

### Cost-effectiveness

In the intervention group, 42 participants completed the EQ-5D-5L questionnaire at both baseline and at the 3-month follow-up. Correspondingly, in the control group, 41 participants described their health using the EQ-5D-5L at both timepoints. Table 1

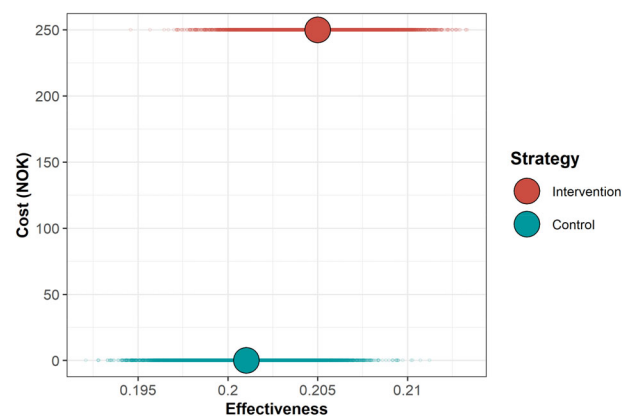


Fig. 2. Bootstrapped trial results. Notes: 10,000 bootstrap replicates of the trial outcomes. The original sample outcomes shown as the larger circles.

Table 1. Group average utility of health, unadjusted QALYs and adjusted QALYs

Group	Average utility (baseline)	Average utility (follow-up)	Unadjusted QALYs	Adjusted QALYs
Control	0.793	0.806	0.1998	0.2010
Intervention	0.822	0.870	0.2114	0.2049
Difference	0.029	0.064	0.0116	0.0039

Note: QALY = quality-adjusted life-year.

presents the calculated average utility for each group at baseline, 3-month follow-up, and the unadjusted average QALYs gained and QALYs gained, adjusted for difference in pre-treatment utility.

**Cost.** The annual operating costs of supplying the intervention was stipulated to 10,000 NOK per year (including both the cost of the system and a limited part time health professional/assistant). Assuming an annual number of users to 40, the per-patient cost of the intervention is 250 NOK per year. As there is no current treatment, the control group cost of zero provides the benchmark with which we compare the cost of the intervention.

**Incremental cost-effectiveness ratio.** Table 2 summarizes the cost-effectiveness analysis. The incremental effectiveness of the intervention was 0.0039 QALYs. The incremental cost per QALYs gained was 63,641 NOK.

**Threshold-value.** The average utility of health in the general Norwegian population between the ages of 0 and 19 is estimated to be 0.93. Between the ages of 12 and 17, the age range of the participants in our study, Statistics Norway estimates an average life expectancy of 71–66 years. This corresponds to between 60 and 55 QALYs. Using the control group's utility of health, our patient group is expected to have between 55 and 51 QALYs remaining. Their absolute short fall of QALYs is therefore approximately four QALYs. According to the prioritization principles, this shortfall would correspond to a cost-effectiveness acceptability threshold of 385,000 in 2015-NOK, or rather 447,000 NOK adjusted for inflation to 2021 (16.1%).

**Likelihood of cost-effectiveness at threshold.** Figure 2 shows the outcome of the non-parametric bootstrapping. Using the percentile method (cutting at the ranked 250th and 9750th iterations), the 95% confidence interval for the average QALYs in the control group was (0.1961, 0.2061). Similarly, in the intervention group, the 95% confidence interval for the average QALYs gained was

Table 2. Incremental results

Group	Cost (NOK)	Effect	Incremental cost	Incremental effect	ICER (NOK/QALY)
Control	0	0.2010	N/A	N/A	
Intervention	250	0.2049	250	0.0039	63,641

Note: ICER = incremental cost-effectiveness ratio; NOK = Norwegian kroner.

(0.2001, 0.2100). Incremental QALYs adjusted for baseline difference was not significant at a 5% level of significance (p-value 0.26). Figure 3 shows the quantification of decision uncertainty at different levels of acceptability thresholds. Beyond a threshold equaling the ICER of 63,641 NOK, the likelihood that the intervention involves a larger net-monetary benefit than no intervention rises with increasing threshold values. At the assumed threshold of 447,000 NOK there is an 83% probability that the intervention will confer an acceptable cost-effectiveness compared to CAU.

## DISCUSSION

The aim of the present study was to investigate whether the web-based YPF intervention could be a cost-effective alternative to offer psychosocial support to adolescents distressed by a visible difference. In short, the results indicated that YPF would be well within what the Norwegian healthcare sector is willing to spend on improvements in QALYs, and thus could be considered a cost-effective alternative to CAU.

YPF is a web-based intervention developed to meet psychological needs among adolescents with distress related to living with a visible difference. The intervention has shown promising results in terms of acceptability (van Dalen *et al.*, 2021; Williamson *et al.*, 2019), effectiveness (Zelihic *et al.*, 2022) and clinical relevance (Kling *et al.*, 2022). However, this study is the first to explore YPF's potential cost-effectiveness, which is important for several reasons. For instance, although cost-effectiveness analyses have increasingly become an important addition to clinical outcome assessment in the evaluation of psychosocial treatments, there is still a general knowledge gap with regard to the costs and cost-effectiveness of intervention and prevention for mental health problems in children and adolescents in most European countries (Kilian, Losert, Park, McDaid & Knapp, 2010). Moreover, YPF is a new intervention addressing a gap (and not aimed to replace an already existing intervention) with the goal of offering easy-accessible and cost-effective psychosocial help to adolescents struggling with living with a visible difference (Williamson, Griffiths & Harcourt, 2015). Interventions can generally be presented as a stepped-care model: whereas most patients benefit from low-level interventions such as information leaflets, more vulnerable individuals require high-level interventions from specialist healthcare services. YPF was developed to support young people in need for more than information, but not requiring complex, face-to-face psychosocial interventions. Hence, in addition to finding out if the intervention works, we also need to examine the program's health economic cost in order to justify its potential implementation. This is especially important considering that we expect few (approx.  $N = 40/\text{year}$ ) to use the program in Norway, based on experiences from the UK and recruitment efforts needed for the Norwegian RCT (Kling, Nordgreen, Kvaem, Williamson & Feragen, 2021). Although the number of expected users might seem a bit low for a web-based intervention, it must be kept in mind that YPF is aimed at a specific population (i.e., adolescents with a visible difference experiencing appearance-related and social distress). However, if the number of users would exceed 40 users per year, this would possibly affect the costs and the health economic

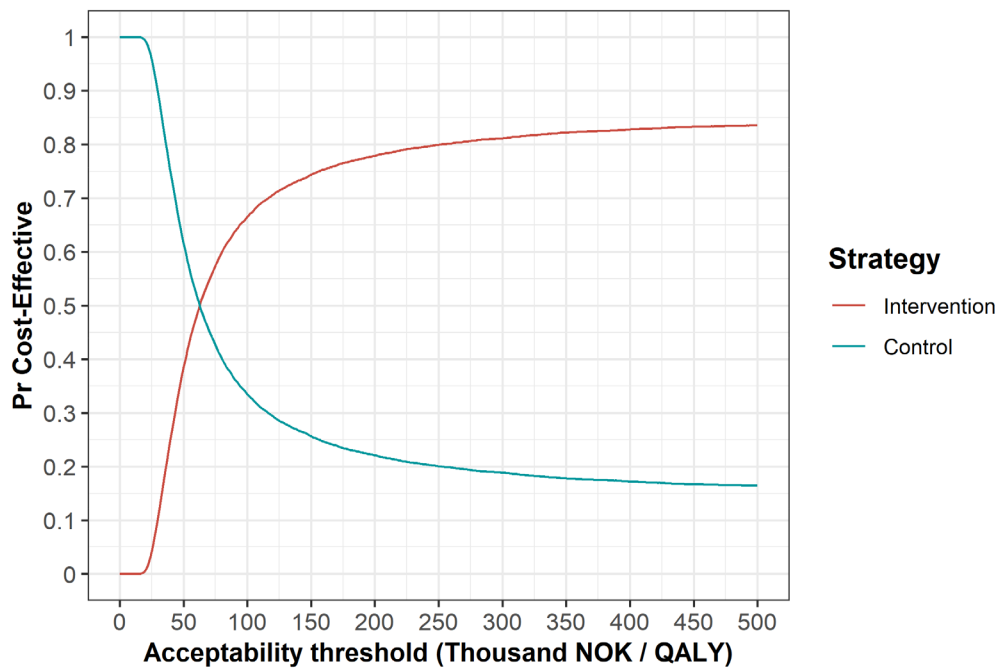


Fig. 3. Cost-effectiveness acceptability curve. Notes: YPF is more likely to be cost-effective than the 'nothing' strategy for a cost-effectiveness threshold exceeding the ICER of 6162.5 NOK. ICER, incremental cost-effectiveness ratio; NOK, Norwegian kroner; YPF, Young Person's Face IT.

assessment of the Norwegian YPF would have to be adjusted accordingly. Specifically, an increased number of users would not change the cost of the program itself, but it would probably increase the costs for salaries to the healthcare professionals and/or assistants involved in YPF support.

It is important to consider that the follow-up in our study was at 3-months. Because of a short-term follow-up, we do not know whether a treatment effect will be sustained beyond this. Hence, our results regard short-term effects of the program and we do not know whether there are longer-term improvements of psychosocial adjustment, or whether improvements decline with time, which is also important to consider in relation to an economic evaluation. For instance, there could be additional long-term health economic benefits associated with the program, such as preventing the need of more extensive psychological/medical treatment, and/or reduced participation in society. Research on anxiety disorders and appearance distress, for example, have shown that early interventions are imperative since, if left untreated, they tend to cumulate depressive symptoms, school/work absence, and social- and economic problems, and that early interventions could reduce distress for the individual and lower the costs to the healthcare system and society (e.g., Arikian & Gorman, 2001; Williamson & Rumsey, 2017). It should be noted that, due to the limited follow-up period of 3 months, potential positive long-term effects of YPF are speculative, as it could also be that differences between intervention and control group diminish over time. Moreover, importantly, it should be acknowledged that using YPF could also induce demands on other psychological resources (e.g., that this intervention would prompt patients to seek further professional care to improve their health further), thereby increasing direct healthcare costs. While the cost of supplying the intervention is arguably low, we do not know to if, or to what extent, it reduces

(or increases) supplemental healthcare resource use. To shed light on this issue, and gain knowledge about the use of YPF and associated costs over time, there is a need for longitudinal research to evaluate both effectiveness and cost-effectiveness aspects of YPF.

This study was performed in Norway, using recommendations for methods and economic thresholds from e.g. the Norwegian Directorate of Health (2012) and the Norwegian Medicines Agency's (2021). Based on other recommendations and other countries' healthcare sectors thresholds for how much to spend on improvements in QALYs, results would differ. However, the conclusion that YPF is likely to be a cost-effective intervention, mainly due to low administrating and personnel costs, is most likely valid also outside of Norway.

#### Limitations and strengths

The present study also has some specific limitations that need to be addressed. First, and as discussed above, the fact that the present study only included a short-term follow-up limits the results and the conclusions that can be drawn. Future studies would benefit from investigating the prospective influence of YPF over more than two time points in order to more efficiently investigate the prospective trajectories, and draw clearer conclusions regarding its potential cost-effectiveness. Second, another major limitation is that the present study included limited cost information beyond the cost of the intervention itself (i.e., operating costs for webhosting YPF, and salaries). As had previously been done in the evaluation of YPF in the UK (Williamson *et al.*, 2019), we sought to collect data on participants' use of healthcare resources, but response rates on this issue were very low and unreliable and thus not included in the present study. Relatedly, clinical experience suggests that this

population generally does not seem to use or find adequate support when struggling with appearance-related distress due to a visible difference, and there is a lack of knowledge regarding if, where and how Norwegian adolescents with a visible difference seek professional psychosocial support. However, it is of importance that future economic evaluations of YPF seek to collect reliable data on resource (healthcare and societal) use in order to investigate whether the intervention results in lower use of services elsewhere in the healthcare sector.

Third, there are issues associated with the used method for calculating improvement (i.e., using a cut-off value). We defined improvement as moving from clinically significant social anxiety pre-intervention to non-clinical/low social anxiety post-intervention, without taking into account the size of the change. However, as the method of defining clinically meaningful change as moving closer to the mean of the functional population than to the mean of the dysfunctional population often is considered the least arbitrary way of calculating clinically significant change (Jacobson & Truax, 1991), this was considered an appropriate way of determining improvement in our sample. Also, the cut-off values for improvement were based on recommendations in the manual (La Greca, 1999), and not values specifically for Norwegian or visible difference populations. However, previous validations in a wide range of different adolescent clinical populations (e.g., Neurofibromatosis and anxiety disorders) have supported the use of the same cut-off values in these groups (La Greca, 1999). Moreover, regarding the measure of health-related quality of life (Norwegian version of EQ-5D-5L; Herdman *et al.*, 2011), it should be acknowledged that this measure was not specifically developed for adolescents and has not been psychometrically evaluated among Norwegian adolescents or adolescents with a visible difference. However, we specifically decided to use the EQ-5D-5L based on national recommendations (Norwegian government, 2016), previous YPF evaluations (Williamson *et al.*, 2019), and since the measure has been found to be valid in other adolescent groups (e.g., Cheung *et al.*, 2016; Welie *et al.*, 2022).

In addition, the cost associated with YPF use in Norway is based on experiences in the UK, and there might be other costs associated with a possible implementation in Norway that we have not foreseen. Likewise, the estimated cost for YPF in Norway could also be exaggerated. Regardless, it is important to interpret the results in light of this uncertainty. Moreover, although we aimed to evaluate the cost-effectiveness analysis of YPF relative to CAU, future studies are encouraged to also compare the intervention to other psychological interventions (e.g., face-to-face) in order to explore its potential relative cost-effectiveness.

Compared to interventions aimed at populations with somatic diseases, generally less is known of the cost-effectiveness of psychological interventions in Norway. As found in this study, we possibly expect there to be short term gains in the utility of health for adolescents distressed by a visible difference. A strength of this study is that we were able to utilize the same approach as for the economic evaluation of somatic diseases. This could contribute to highlighting the importance of also prioritising mental health within the health care system.

## Conclusion

Psychological interventions aimed at adolescents with a visible difference and experiencing psychosocial distress are lacking. The web-based intervention YPF was developed to address this gap and to serve as an easy-accessible and cost-effective intervention. The results of the present study indicate that YPF is well within acceptability thresholds for costs in the Norwegian healthcare sector. However, more research is needed in order to determine the intervention's potential long-term cost-effectiveness, as well as the potential of the intervention to reduce (or increase) demands on other health- and societal resources.

This study was funded by the Norwegian Research Council (Grant number: 287243). Clinical trial registration: NCT03165331. The trial has been reviewed by the Regional Committee for Medical Research Ethics (Health Region South-East, reference number: 2015/2440), and accepted by the hospital's Data Protection Office.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request. R code is available in the Supplementary Appendix.

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#### SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article:

#### Appendix S1. Supplementary Appendix.

Received 10 June 2022, Revised 19 October 2022, accepted 24 October 2022