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1 Psychosocial interventions for adults with visible differences: A systematic review

2 Running title: Adults and visible difference review

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Abstract

Background: Some individuals with visible differences have been found to experience psychosocial adjustment problems that can lead to social anxiety and isolation. Various models of psychosocial intervention have been used to reduce social anxiety and appearance related distress in this population. The objective of this review was to update a previous systematic review assessing the efficacy of psychosocial intervention programs for adults with visible differences. The original review (Bessell & Moss, 2007) identified 12 papers for inclusion.

Methods: A search protocol identified studies from 13 electronic journal databases.

Methods: Studies were selected in accordance with pre-set inclusion criteria and relevant data were extracted.

Results: This update identified an additional four papers that met the inclusion criteria. Two papers provided very limited evidence for the efficacy of a combined cognitive-behavioural and social skills training approach. None of the papers provided sufficient evidence for the optimal duration, intensity or setting of psychosocial interventions for this population.

Discussion: The review concluded that a greater number of Randomised Controlled Trials and experimental studies were required to increase the methodological validity of intervention studies.

Keywords:

- Visible differences, Psychosocial, narrative synthesis, Cognitive-behavioural therapy, Social skills training

36 Introduction

37 The term visible difference refers to any kind of condition, whether congenital or acquired
38 that can leave an individual with an altered appearance (e.g. skin conditions, burns, scarring
39 or craniofacial abnormalities. Some individuals with visible differences have been found to
40 experience psychosocial adjustment problems that can lead to social anxiety and isolation
41 (Rumsey et al, 2004; Rumsey & Harcourt, 2012) and poor quality of life (Marcusson, Paulin &
42 Ostrup, 2002). As such, appearance altering conditions present a clear challenge to a
43 positive body image for those affected and have led to the development of numerous
44 psychosocial intervention programs designed to address the psychological, as well as the
45 physical needs and difficulties experienced by those with visible differences. The
46 psychosocial difficulties experienced by some of those with visible differences include name
47 calling, staring and unsolicited questioning about their appearance (Kleve & Robinson,
48 1999).

49 There are many different models that outline the difficulties experienced by some
50 individuals with visible differences. These include the social anxiety model (Baumeister &
51 Leary, 1995), Goffman's (1968) model of stigma, social skills models (Bull & Rumsey, 1988)
52 and models of body image disturbance (Cash, 2001). Baumeister and Leary's (1995) model
53 suggests that individuals with visible differences experience social anxiety at least in part
54 because they are fearful of being rejected or excluded on the grounds of having an unusual
55 or different appearance (Kent, 2000). Therefore, this model suggests that it is important to
56 focus interventions on reducing social anxiety through exposure to social situations in order
57 to promote positive adjustment amongst those with visible differences (Newell & Marks,
58 2000). Goffman's (1968) stigma model fits in many ways with the social anxiety model, and
59 states that having a different appearance is a characteristic that is "devalued" by society and

60 as such those with a visible difference are more likely to be excluded or rejected, which
61 suggests a very real reason for experiencing social anxiety.

62 Some research has suggested that those with visible differences can become
63 preoccupied with their own appearance due to high levels of distress (Clarke, 1999). This
64 preoccupation can make people seem distracted or lacking confidence when they are in
65 public (Kent, 2000). Therefore, the social skills model suggests that many of the negative
66 reactions that they experience from others are less to do with stigma, as Goffman's (1968)
67 model would suggest, but more a reaction to the poorer social skills that the person with
68 the visible differences is exhibiting (Bull & Rumsey, 1988). These two models do not
69 necessarily have to be mutually exclusive. The reality of the situation for many people with
70 visible differences is indeed that they experience some level of rejection and exclusion from
71 others, but in some cases this effect is exacerbated by the poor social skills that they have
72 developed (Kent, 2000). Therefore, focusing on improving social skills is a key focus for
73 intervention models (Rumsey, Robinson & Partridge, 1993).

74 Finally, the body image disturbance model (Cash, 1996) suggests that in the case of
75 visible difference, the individuals may experience dissatisfaction with their body image
76 because they do not conform to the cultural norms of attractiveness that their society
77 imposes. This social pressure to look a certain way, alongside a more personal form of
78 stigma, where they themselves feel they should look "normal", can lead to high levels of
79 body image disturbance, which is associated with poorer adjustment (Altabe & Thompson,
80 1996). This model suggests that interventions should focus specifically on addressing the
81 way individuals feel about their appearance and the negative assumptions they make about
82 the importance of appearance.

83 The reality is that no one model completely explains the experience of living with a
84 visible difference. Kent (2000) recommended an integrated model that addresses body
85 image dissatisfaction and the negative assumptions associated with appearance concerns.
86 He also suggested that it is important to target social anxiety with exposure therapy
87 (introducing people to feared social situations). However, as there is a very real tendency for
88 individuals to experience negative responses from others, it is important to boost social
89 skills too, in order to provide individuals with the techniques that they will need to deal with
90 these responses. Both social skills training (SST) and cognitive behavioural therapy (CBT) are
91 common intervention types for adults with visible differences.

92 Although these intervention techniques for people with a visible difference are used,
93 there is still a significant lack of evidence pertaining to the efficacy of these different
94 psychosocial techniques. A systematic review conducted by Bessell and Moss (2007) found
95 little to no evidence to support any particular intervention model, due to methodological
96 constraints associated with the included studies. Since the review was published other
97 studies have assessed the efficacy of various psychosocial intervention models for adults
98 with visible differences. For that reason it is important that the original review be updated
99 to ensure an accurate evidence base for psychosocial interventions for this population.

100 A recent systematic review conducted by Muftin and Thomson (2013) looked at self-
101 help psychosocial interventions for individuals with visible differences. Whilst this is an
102 important update, the review does not incorporate all forms of psychosocial intervention,
103 only those administered in a self-help format. Therefore the review does not help to answer
104 fundamental questions raised by our original review regarding method of delivery (Bessell &
105 Moss, 2007). It is therefore, the belief of the current authors that this update is both needed
106 and timely.

107 Objectives

108 The aim of the present study was to update the existing systematic review (Bessell & Moss,
109 2007) of the efficacy of psychosocial intervention programs for adults with visible
110 differences from 2006 (the date of the last search) to the present day. Where appropriate,
111 meta-analysis was used to synthesise findings across papers. The overall intention of this
112 study was to identify methodological issues in need of further attention in this area of
113 research. Furthermore, we hoped that this review would aid in the development of new
114 intervention programs within the field of visible differences.

115 Materials & Methods

116 *Study selection*

117 The search aimed to identify all studies relating to psychosocial interventions for adults with
118 visible differences from January 2006 (six months prior to the original search in Bessell &
119 Moss, 2007) to 12th May 2014. An extensive search strategy was used to search 13
120 databases, including Medline, embase, psychinfo, and Cochrane central register of
121 Controlled trials (CENTRAL) (See Appendix A for full search strategy). This was compiled by a
122 library technician based on an exhaustive list of appearance altering conditions and types of
123 psychosocial intervention. No language restrictions were applied. In addition websites
124 including National Institute of Clinical Excellence (NICE) and the metaRegister of Controlled
125 Trials (mRCT) were searched and reference lists of included papers. Search criteria were
126 adapted to suit the search terms of each individual database.

127

128

129

130 *Inclusion criteria*

131 Study design: No exclusions were applied based on study design with all study designs being
132 included in the review. Case studies with less than five participants in each group were
133 excluded.

134 Population: Adults with noticeable visible differences, e.g. disfigurements of face, neck and
135 hands. This included a wide range of different conditions from congenital skin conditions
136 and abnormalities to cancer patients, or those with scars resulting from injury. All client
137 groups were over the age of 16. Both males and females of any ethnicity or race were
138 included. Any study containing less than 90% adults with visible differences where data
139 were not provided separately for those individuals were excluded from the review unless
140 the data were available from the authors.

141 Interventions: These included CBT, SST and more traditional forms of psychotherapy all
142 delivered either alone or as part of a package of care. The interventions had to include some
143 element specifically designed to target appearance concerns.

144 Comparators: The comparators used in this review were current standard treatments
145 including standard therapist-led CBT for the treatment of anxiety or depression, non-
146 directive counselling, primary care counselling, routine management (drug treatments for
147 anxiety or depression) and no treatment.

148 Outcomes: The primary outcome measure was any measure of appearance related distress
149 (e.g. body image concerns, body image quality of life etc.). Only studies with this primary
150 outcome measure were included in the review¹. Secondary measures included measure of
151 anxiety and depression and general improvements in psychological symptoms, interpersonal

¹ The aim of this review was specifically to assess appearance-related distress not general psychosocial functioning and differs to other reviews, e.g. Muftin & Thompson, 2013.

and social functioning, satisfaction and preference, site of delivery and acceptability of treatment.

Exclusion Criteria

Any treatment designed to treat dysmorphophobia, body dysmorphic disorder or eating disorders such as bulimia nervosa or anorexia nervosa were removed. It was also decided to exclude any visible differences that were not considered to be commonly on display (such as breast reconstruction, abdominal injury), due to the vast amounts of literature available on these conditions. These types of conditions do fall within the remit of visible differences, but it was considered that the needs of individuals with “hidden” differences might be different to those with normally visible differences, meaning that different intervention techniques may be appropriate.

Ethical considerations

As this review is concerned with the analysis of published data, ethical considerations regarding direct contact with participants were not applicable.

Analysis

The authors used a qualitative approach to synthesise data across studies (Dixon-Woods et al., 2005) and focused on three main areas: information pertaining to theoretical or therapeutic perspective, method of delivery (setting, person delivering the intervention) and timing of the intervention (intensity and frequency of the intervention).

175 Meta-analysis of Trials only

176 Outcome Measures: Primary and secondary outcome measures of psychosocial adjustment
177 were extracted (e.g. preoccupation with appearance, anxiety, depression, confidence,
178 quality of life, social integration).

179 Effect Sizes: Standard mean differences (SMDs) and/or effect sizes together with 95%
180 confidence intervals (CIs) were extracted for continuous outcomes and odds ratios (ORs)
181 together with 95% CIs were extracted for dichotomous outcomes. These figures were taken
182 directly from the papers or calculated based upon raw data provided within the papers
183 where necessary. Effect sizes and confidence intervals were plotted using forest plots. Meta
184 analyses were only conducted where multiple studies were randomised controlled trials
185 (RCTs) with similar interventions to allow appropriate data pooling.

186 Assessment of risk of bias

187 Three reviewers (AN, AM & JG) independently assessed trials using the Cochrane Risk of Bias
188 tool (Higgins & Green, 2011) to rate each of the following five components as high, low or
189 unclear risk of bias: 1) method of sequence generation, 2) method of allocation
190 concealment, 3) method of blinding of the outcome assessor, 4) selective reporting of
191 outcome data (reporting all outcomes in the results that are mentioned in the method and
192 using standard outcome measures within a particular field of research) and 5) completeness
193 of outcome data (attrition rates and intention to treat (ITT) analyses).

194 In the case of observational studies two reviewers (AN & JG) used the RAMbo
195 assessment tool (Chen & Wang, 2009) to assess the quality of randomization (R), whether
196 missing data was accounted for (A) and whether the type of measurement was appropriate
197 (M).

198

Results & Discussion

The search results identified 13837 possible studies since the previous review was conducted. After removal of duplicates, 3539 studies were identified for further investigation. Of these 3468 studies were identified as not relevant for inclusion within the review and were discarded on the basis of titles and abstracts independently by at least two reviewers (AN, AM & JG). This left 71 studies to assess for inclusion (See figure 1).

Insert figure 1 here

Of the 71 papers identified for possible inclusion, four met the inclusion criteria on closer inspection by three reviewers (AN, TM & AM). Sixty-seven studies were excluded. Reasons for exclusion included studies that did not assess an intervention targeting appearance or related psychosocial distress (27 studies), those that did not assess an intervention (five studies), case studies with less than 5 participants in each group (13 studies), descriptive articles or review papers (14 papers), those with no primary outcome measure of appearance-related distress or body image concern (6 studies) and two which met the inclusion criteria, but not enough data was present in the abstracts to include within the review (authors were contacted for full papers but were not supplied).

Risk of Bias Assessment

Two papers (Srivastava & Chaudhury, 2014; Bessell et al, 2012); were assessed using the Cochrane risk of bias assessment tool which is suitable for assessing RCTs (Higgins & Green, 2011). The Bessell et al (2012) paper was assessed for risk of bias by two researchers independent of the paper's authors (AM & JG) as two of the authors were also the authors of this review.

221 Risk of Bias Assessment: Of the two papers, one (Bessell et al 2012) was found to be of low
 222 risk of bias with regards to randomization sequence and allocation concealment (See table
 223 1). Only one paper was found to have low risk of bias for blinding of outcome assessor
 224 (Bessell et al 2012). All rates of attrition were adequately documented in the papers.
 225 Srivastava and Chaudhury (2014) did not report any attrition rates throughout the study
 226 period. All outcomes reported in the studies were reported in the results.

227 *Insert Table 1 here*

228 RAMbo Assessment: Two papers (Jolly et al, 2010; Semple, Dunwoody, Kernohan &
 229 McCaughan, 2009) were assessed using the RAMbo technique for observational studies (see
 230 Table 2). Jolly et al (2010) did not report using a randomisation procedure, so was rated as
 231 unclear, whilst Semple et al, 2009 did not use a randomisation technique so was rated at
 232 high risk of bias. Semple et al (2009) was rated at low risk of bias for attrition and
 233 measurement, whereas Jolly et al (2010) was rated as unclear as multiple abstract
 234 publications of this study refer to different numbers of participants. The study was also
 235 rated unclear for measurement as results for the anxiety outcome measure were not
 236 reported.

237 *Insert table 2 here*

238 Effects of Interventions: Therapeutic approach

239 Cognitive-Behavioural Therapy: Jolly et al (2010) assessed the efficacy of an individual CBT
 240 program for patients with lupus. The intervention focused on body image education, self-
 241 esteem, anxiety and depression and also contained cosmetic training. The study employed
 242 15 women with lupus (10 treatment and 5 controls) through a clinic in the United States.
 243 The mean ages of the participants in the treatment and control groups were 43.6 years and
 244 39.3 years respectively. Outcome measures included Multi-Dimensional Body Relations

245 Satisfaction – Appearance Scale (MBRSQ-AS), Situational Inventory of Body Image Dysphoria
 246 (SIBID-SF), Body Image in Lupus Screen (BILS) and Anxiety and Lupus PRO (Table 3).

247 *Insert Table 3 here*

248 The previous review by Bessell and Moss (2007) did not include meta-analyses. The authors
 249 of the current review revisited the data from previous papers with a view to conducting
 250 meta-analyses on any studies that consisted of randomised trials. Two of the original papers
 251 met this criterion (Papadopoulos, Walker & Anthis, 2004; Newell & Clarke 2000). The Newell
 252 and Clarke (2000) paper did not contain sufficient detail to allow a meta-analysis to be
 253 conducted. No other CBT studies consisted of randomised trials, so it was not possible to
 254 conduct a meta-analysis on this intervention type. Overall the review concluded there was
 255 very limited evidence for the efficacy of CBT for adults with visible differences.

256 Combined CBT and SST: Bessell et al (2012) assessed the efficacy of two psychosocial
 257 interventions against a no-treatment control. The first intervention consisted of a face-to-
 258 face CBT/ SST intervention, whilst the second was an online delivery of the same
 259 intervention model. The study employed 83 individuals with varying visible differences
 260 recruited through charity organizations, the Royal Free Hospital, London outpatient plastic
 261 surgery clinic and general advertising. Participants (34 male, 49 female) were over 18 years
 262 of age, with a mean age of 45 years (see Table 2 for study information). Outcome measures
 263 used included the Hospital Anxiety and Depressions Scales (HADS), the Derriford
 264 Appearance Scale-24 (DAS-24), and the Body Image Quality of life Inventory (BIQLI).

265 Semple et al (2009) assessed the efficacy of an individual CBT/ SST program for patients with
 266 head and neck cancer. The intervention focused on a series of specific areas including
 267 anxiety, depression, fatigue, appearance and stress. The study employed 54 patients with
 268 head and neck cancer recruited through the Regional head and Neck service in Northern

269 Ireland. Participants (40 males, 14 females) were 31 to 75+ years of age. Outcome
270 measures included the HADs, the Work and Social Adjustment (WASA) scale and a health-
271 related quality of life measure (University of Washington quality of life scale version 4)
272 which contained a measure of appearance-related distress.

273 The Semple et al (2009) paper did not contain sufficient detail to allow a meta-analysis to be
274 conducted. No other CBT studies consisted of randomised trials, so it was not possible to
275 conduct a meta-analysis on this intervention type. Overall the review found only very
276 limited evidence for the efficacy of a combined CBT and SST approach for adults with visible
277 differences.

278 Person-centred: Srivastava and Chaudhury (2014) compared treatment as usual (one
279 counselling session; 83 participants) against a six session psychotherapeutic program (90
280 participants). Participants were aged 22 – 52 years of age with a mean age of 30.05. All
281 patients had experienced amputation. Intervention consisted of six session based on
282 reassurance, ventilation of emotions, acceptance of self, therapeutic milieu and
283 reintegration.

284 A study previously cited in the Bessell and Moss (2007) review also assessed the efficacy of a
285 person-centred approach (Papadopoulos et al, 2004). However this study did not contain
286 enough information to allow a meta-analysis to be conducted. Overall this review has found
287 little evidence for the use of the person-centred approach to therapy.

288 Effects of Interventions: Method of Delivery

289 Self-help: One of the included studies assessed the efficacy of self-help interventions. The
290 Bessell et al (2012) paper compared face-to-face delivery of a CBT intervention against an
291 online delivery with minimal facilitation from an assistant psychologist or counsellor.

Face-to-face individual: All four studies assessed the efficacy of individual CBT-based interventions. The Bessell et al (2012) paper also assessed the efficacy of a face-to-face delivery of a CBT/ SST intervention administered by a trained counsellor or an assistant psychologist. The Semple et al (2009) paper assessed a face-to-face CBT/ SST intervention administered by a trained clinical nurse specialist. Jolly et al (2010) assessed the efficacy of individual CBT-based support for women with lupus. Srivastava and Chaudhury (2014) assessed the efficacy of individual psychotherapy delivered by a psychiatric nurse for individuals with amputations.

Due to the differences in methodological design, it was difficult to draw any firm conclusions about the optimal delivery of psychosocial interventions. Therefore, the review cannot recommend whether any particular individuals should be responsible for delivering these psychosocial interventions.

Effects of Interventions: Timing of Intervention

This review attempted to identify the optimal duration and intensity of intervention. The studies included within this review varied in duration from two sessions (Semple et al, 2009), through to 10 sessions (Jolly et al, 2010). Full details of intervention duration can be found in Table 3. The intensity of the interventions consisted of weekly (Srivastava & Chaudhury, 2014; Bessell et al, 2012) or fortnightly sessions (Semple et al, 2009). Sessions were between one and two hours in length (see Table 3 for full details of intensity).

Due to the differing intensity and duration across the studies, it is difficult to draw any firm conclusions regarding the optimal length and intensity of therapy. However, most studies opted for between 6 - 10 sessions administered weekly for 1-1.5 hours. Therefore, it would seem reasonable to conclude that this is the minimum intensity and duration required to

316 lead to clinically significant changes in appearance-related distress and anxiety. This also
317 matches recommendations for the minimum intensity of therapies in the general population
318 (Roth & Fonagy, 2005).

319 Effects of Interventions: Participant Acceptability

320 As well as assessing efficacy of interventions, it is important that trials of interventions also
321 measure patient acceptability. One paper reported on overall acceptability (Bessell et al,
322 2012; Newell & Clark, 2000). The Bessell et al (2012) provided information about overall
323 acceptability, as well as ratings of usefulness and satisfaction for both the face-to-face and
324 computer-based intervention. Users of the face-to-face intervention gave it an average
325 usefulness rating of 8.23 out 10 and a satisfaction rating of 8 out 10. The computer
326 intervention was given ratings of 8.79 and 8.38 out of 10 respectively. Overall acceptability
327 for the face-to-face intervention was 51.89 out of 60 and 52.7 out 60 for the computer
328 intervention. The original Bessell and Moss (2007) review also included a study by Newell
329 and Clarke (2000) which measured patient acceptability (not included in the previous
330 review). Newell and Clark (2000) paper found that 68.75% found the leaflet useful. Only
331 9.38% rated the booklet as unhelpful. These papers suggest that the CBT or combine CBT
332 and SST approach may be viewed as acceptable by adults with visible differences.

333 *Main findings*

334 The strength of the evidence to support the efficacy of the existing interventions from this
335 narrative synthesis is generally poor. The methodological quality of the included studies was
336 limited and small intervention effect sizes were observed. The studies looked at differing
337 interventions making judgments about consistency across studies difficult because each
338 study used different intervention settings, e.g. group, self-help or face-to-face and
339 paradigms, e.g. CBT, SST or person-centred. There is some very limited evidence to support

340 the efficacy of a combined CBT and SST approach to support, but this is far from conclusive
341 as it is based on a combined sample size of 137 participants.

342 The length of intervention required was unclear with studies ranging from six to 10
343 sessions. No firm conclusions can be made regarding the optimum therapy time required to
344 reduce psychosocial difficulties, or the most appropriate setting for these interventions.
345 Neither can conclusions be drawn about the level of therapist contact or expertise required
346 to produce optimum results. Due to the wide-ranging use of therapeutic paradigms of each
347 intervention, it was not possible to draw any firm conclusions regarding the acceptable
348 content of psychosocial interventions for the visibly different population, or the adequate
349 implementation of these interventions. The participant populations were also varied in
350 terms of conditions and symptom severity. Further studies need to be conducted to
351 establish which interventions are most effective for specific sub-populations.

352

353 *Interpretation of findings in relation to previously published work*

354 The findings of this review were no different to the conclusions of the original review
355 (Bessell & Moss, 2007), which made recommendations for a greater number of future
356 studies, including more RCTs and experimental studies. Furthermore the need for greater
357 methodological vigour was highlighted with regards to ITT analyses, greater detail pertaining
358 to attrition characteristics, rates and causes, greater sample sizes, clearer inclusion and
359 exclusion criteria, and studies that measure interventions against control groups as
360 standard. The review also emphasized the need for patient acceptability ratings.

361 Seven years on from the publication of the original review and it would appear that little
362 has changed within this research field. The authors of this update decided to use a tighter
363 inclusion criteria than used previously to ensure only studies that measured body image or

364 appearance-related distress were included within the analysis. This limited the number of
365 new studies to just four. This highlights a desperate need for more research within this area,
366 with studies measuring body image and/or appearance-related distress as standard.
367 Furthermore, of the four new studies included in this update, only one consisted of a RCT
368 reported in sufficient detail for low risk of bias and suitable for data pooling (Bessell et al,
369 2012). As this study was conducted by the two of the authors of this review demonstrates
370 how important this timely update is for reminding future researchers of the importance of
371 rigorous experimental design.

372 Current practice involves very limited testing of the efficacy of interventions, and this
373 needs to be addressed. Within the UK, the lack of service provision within the NHS has led
374 to an increased need amongst this population (Bessell et al, 2010). The authors suggest that
375 the reason for the lack of scientifically tested interventions is that many self-funded
376 charities have had to pick up the shortfall in service provision and these organizations have
377 been more concerned with spending money on providing services than on evaluating them.
378 Furthermore, with limited money available for research into visible difference, research
379 centres are hard pushed to carry out cheap and quick evaluations whilst ensuring scientific
380 rigor does not suffer. The resources involved in performing fully blind RCTs for psychosocial
381 interventions are expensive and require large clinical and research team, which most
382 budgets do not allow for.

383

384

385 *Strengths and Limitations of this study*

386 Credit must be given to the existing studies for trying to evaluate interventions for such a
387 hard-to-reach population. Designing interventions specifically for certain conditions

classified as affecting appearance can be very difficult due to the rarity of some conditions. Even when designing interventions for a wide range of conditions, the population can still be difficult to reach leading to low sample sizes and the population can vary widely, making generalizability a problem. Therefore this review was based on small populations and meta-analysis was not possible due to differences in study design. Future research needs to consider the use of multi-site studies in order to recruit larger numbers of participants and thus increase the reliability of the findings of such evaluations.

Implications for future research, policy and practice

It must be emphasized that despite the methodological problems associated with assessing these interventions, the techniques themselves are still important. Although their efficacy still needs further establishment, these interventions are necessary for increasing service provision for individuals with visible differences. These include interventions run by the specialist psychological outpatient clinic at Frenchay Hospital in Bristol, UK, the UK charity Changing Faces, the Face IT online tool, and other techniques in the US, such as the social skills interventions run by Kathy Kapp-Simon for adolescents with cleft lip and palate through the charity AboutFace USA in Illinois, and those run by Pat Blakeney for those with burns injuries at Galveston Burns Hospital in Texas. They are also needed to address the issue of an overall package of care for visibly different clients from medical treatment right through to adjustment and psychosocial functioning. For these reasons, further testing of these interventions is a fundamental step.

The current interventions have provided very limited support for the CBT and combined CBT and SST models. These techniques offer individuals practical solutions to some of their social difficulties without pathologising them. Although it is clear that there is a need for

412 individuals to have access to resources such as grief or trauma counselling, particularly after
413 an acquired difference in order to cope with changes in body image, many individuals simply
414 require brief solution-focused interventions. This can be provided by CBT and SST
415 techniques. Furthermore, evidence from the acceptability measures used in some of the
416 studies that involved these approaches has suggested that individuals with visible
417 differences do find these types of interventions acceptable (Bessell et al, 2012; Newell &
418 Clarke, 2000). This is further supported by a felt needs assessment recently conducted with
419 potential service users within the field of visible difference, which identified that most
420 service users found the idea of CBT or SST to be acceptable and positive (Bessell et al, 2010).
421 This is an interesting point to note as it demonstrates that individuals with visible
422 differences do not find the idea of interventions associated with their appearance
423 stigmatizing, as has often been a concern by experts in the past.

424

425 *Conclusion*

426 Overall this review concludes that to date there is very limited evidence to support the
427 efficacy of CBT or a combined CBT and SST approach for supporting adults with visible
428 differences. However, there is still insufficient information to draw firm conclusions and
429 little to no information available regarding the optimal setting for interventions of this
430 nature, the optimal service provider, length of time or intensity of intervention. All these
431 factors must be addressed in order to demonstrate efficacy in the future. The authors
432 conclude that little has changed in the research community since the publication of the
433 initial review. It is important that future research follows the recommendations made within
434 these reviews.

435

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439

440 References

441 Altabe, M., & Thompson, J.K. (1996). Body Image: A cognitive self-schema construct.

442 *Cognitive Therapy and Research*, 20, 171-193.

443 Baumeister, R.F., & Leary, M.R. (1995). The need to belong: Desire for interpersonal

444 attachments as a fundamental human motivation. *Psychological Bulletin*, 117, 497-

445 529.

446 Bessell, A., Brough, V., Clarke, A., Harcourt D., Moss T., & Rumsey, N. (2012). Evaluation of

447 the effectiveness of Face IT, a computer-based psychosocial intervention for

448 disfigurement-related distress. *Psychology, Health and Medicine*, 5, 565-577.

449 Bessell, A., Clarke, A., Harcourt, D., Moss, T.P., & Rumsey, N. (2010). Incorporating user

450 perspectives in the design of an online intervention tool for people with visible

451 differences: Face IT. *Behavioural and Cognitive psychotherapy*, 38, 577-596.

452 Bessell, A., & Moss, T.P. (2007). Psychosocial interventions for visible differences: A

453 systematic review. *Body Image*, 4, 227-238.

454 Bull, R., & Rumsey, N. (1988). *The social psychology of facial appearance*. New York: Springer

455 Verlag.

456 Cash, T.F. (2001). *The body image workbook*. Oakland CA: New Harbinger Publications.

- 457 Cash, T.F. (1996). The treatment of body image disturbances. In K. Thompson (Ed.), *Body*
 458 *image, eating disorders and obesity*. Washington, DC: American Psychological
 459 Association.
- 460 Chen, J.F., & Wang, C.F. (2009). Introduction of critical appraisal of medical literature.
 461 *Medical Dispute Mediation*, 42, 19-24.
- 462 Clarke, A. (1999). *Everybody's staring at me!* London: Changing Faces.
- 463 Dixon-Woods, M., Agarwal, S., Jones, D., Young, B., & Sutton, A. (2005). Synthesising
 464 qualitative and quantitative evidence: a review of possible methods. *Journal of*
 465 *Health Services Research & Policy*, 10:45-53.
- 466 Goffman, E. (1968). *Stigma*. London: Penguin books
- 467 Higgins, J.P.T., & Green, S. (2011). *Cochrane handbook for systematic reviews of*
 468 *interventions*. Oxford: The Cochrane Collaboration.
- 469 Jolly, M., Peters, K., Cash, T.F., Mikolaitis, R.A., Gao, W., & Block, J.A. (2010). *Body Image*
 470 *intervention improves health outcomes in systemic Lupus Erythematosus*. Poster
 471 session presented at the Lupus Conference: 9th International Congress, Vancouver,
 472 CA.
- 473 Kent, G. (2000). Understanding experiences of people with disfigurement: An integration of
 474 four models of social and psychological functioning. *Psychology, Health and*
 475 *Medicine*, 5, 117-129.
- 476 Kleve, L., & Robinson, E. (1999). A survey of psychological needs in adult burn-injured
 477 patients. *Burns: Journal of the International Society for Burn Injuries*, 25, 575-579.
- 478 Marcusson, A., Paulin, G., & Ostrup, L. (2002). Facial appearance in adults who had cleft lip
 479 and palate treated in childhood. *Scandinavian Journal of Plastic and Reconstructive*
 480 *Surgery and Hand Surgery*, 36, 16-23.

- 481 Muftin, Z., & Thompson, A.R. (2013). A systematic review of self-help for disfigurement:
482 Effectiveness, usability and acceptability. *Body Image*, 10, 442-450.
- 483 Newell, R., & Clarke, M. (2000). Evaluation of a self-help leaflet in treatment of social
484 difficulties following facial disfigurement. *International Journal of Nursing Studies*,
485 37, 381-388.
- 486 Newell, R., & Marks, I. (2000). Phobic nature of social difficulty in facially disfigured people.
487 *British Journal of Psychiatry*, 176, 177-181.
- 488 Papadopoulos, L., Walker, C., & Anthis, L. (2004). Living with vitiligo: A controlled
489 investigation into the effects of group cognitive-behavioural and person-centred
490 therapies. *Dermatology and Psychosomatics*, 5, 172-177.
- 491 Roth, A., & Fonagy, P. (2005). *What works for whom? (2nd ed): A critical review of*
492 *psychotherapy research*. New York: The Guilford Press.
- 493 Rumsey, N., Clarke, A., White, P., Wyn-Williams, M., & Garlick, W. (2004). Altered body
494 image: Auditing the appearance related concerns of people with visible
495 disfigurement. *Journal of Advanced Nursing*, 48, 443-453.
- 496 Rumsey, N., & Harcourt, D. (2012). *The Oxford handbook of the psychology of appearance*.
497 Oxford: Oxford University Press.
- 498 Rumsey, N., Robinson, E., & Partridge, J. (1993). *An evaluation of the impact of social skills*
499 *training for facially disfigured people*. Bristol: Changing Faces.
- 500 Semple, C., Dunwoody, L., Kernohan, W.G., McCaughan, E. (2009). Development and
501 evaluation of a problem-focused psychosocial intervention for patients with head
502 and neck cancer. *Support Care Cancer*, 17(4), 379-388.

503 Srivastava, K., & Chaudhury, S. (2014). Rehabilitation after amputation: Psychotherapeutic
504 intervention module in Indian scenario. *Scientific World Journal*, doi:
505 10.1155/2014/469385.
506

507 Appendix A: Sample search strategy
 508 1 exp Adaptation, Psychological/
 509 2 exp Psychotherapy/
 510 3 exp Counseling/
 511 4 "Self-Help Groups"/
 512 5 "Social Support"/
 513 6 ((psychosocial\$ or psycho-social\$) adj5 (intervention\$ or treatment\$ or therap\$ or program\$)).tw.
 514 7 counsel\$.tw.
 515 8 (behavi\$ adj5 (therap\$ or treatment\$ or program\$ or intervention\$)).tw.
 516 9 (cognitiv\$ adj5 (therap\$ or treatment\$ or program\$ or intervention\$)).tw.
 517 10 (psychologic\$ adj5 (therap\$ or treatment\$ or program\$ or intervention\$)).tw.
 518 11 (mindfulness adj5 (therap\$ or treatment\$ or program\$ or intervention\$)).tw.
 519 12 "Early Intervention (Education)"/
 520 13 Patient Education as Topic/
 521 14 support group\$.tw.
 522 15 self-help.tw.
 523 16 psychotherap\$.tw.
 524 17 group therap\$.tw.
 525 18 Social Adjustment/
 526 19 person-cent\$ therap\$.tw.
 527 20 solution-based therap\$.tw.
 528 21 or/ 1-20
 529 22 exp Cicatrix/
 530 23 ((face or facial) adj3 scar\$).tw.
 531 24 (visible adj3 scar\$).tw.
 532 25 keloid\$.tw.
 533 26 cicatrix.tw.
 534 27 exp Facial Injuries/
 535 28 ((facial\$ or face) adj3 (injur\$ or damage\$)).tw.
 536 29 exp Craniofacial Abnormalities/
 537 30 exp Facial Dermatoses/
 538 31 facial dermatos\$.tw.
 539 32 Psoriasis/
 540 33 psoriasis.tw.
 541 34 Eczema/
 542 35 eczema.tw.
 543 36 exp Skin Abnormalities/
 544 37 Epidermolysis Bullosa.tw.
 545 38 port wine stain\$.tw.
 546 39 exp Hemangioma/
 547 40 h?emangioma\$.tw.
 548 41 exp Pigmentation Disorders/
 549 42 vitiligo.tw.
 550 43 exp "nevi and melanomas"/
 551 44 (birth mark\$ or birthmark\$).tw.
 552 45 melanoma\$.tw.
 553 46 burns/
 554 47 burns.tw.
 555 48 exp Alopecia/
 556 49 alopecia.tw.
 557 50 exp Exophthalmos/
 558 51 exophthalm\$.tw.
 559 52 thyroid eye disease.tw.
 560 53 exp Strabismus/
 561 54 strabismus.tw.
 562 55 (misalign\$ adj3 eye\$).tw.

563 56 exp Eyelid Diseases/
 564 57 exp Mouth Neoplasms/
 565 58 ((mouth or oral) adj3 (neoplasm\$ or cancer\$ or tumor\$)).tw.
 566 59 Growth Disorders/
 567 60 exp Dwarfism/
 568 61 Gigantism/
 569 62 dwarfism.tw.
 570 63 dwarf.tw.
 571 64 small stature.tw.
 572 65 gigantism.tw.
 573 66 restricted growth.tw.
 574 67 exp Dystonia/
 575 68 Torticollis.tw.
 576 69 dystonia.tw.
 577 70 Dupuytren Contracture/
 578 71 Dupuytren\$ contracture\$.tw.
 579 72 Amputation/
 580 73 Artificial Limbs/
 581 74 Amputees/
 582 75 amputees.tw.
 583 76 artificial limb\$.tw.
 584 77 (appearance adj5 (abnormal or malformation or problem\$)).tw.
 585 78 (visibl\$ adj5 disabilit\$).tw.
 586 79 (visibl\$ adj5 differen\$).tw.
 587 80 disfigur\$.tw.
 588 81 (appearance adj5 (malform\$ or problem\$)).tw.
 589 82 (deformit\$ or deformed).tw.
 590 83 (appearance\$ adj5 (distress or anxiety or depression)).tw.
 591 84 appearance.ti.
 592 85 exp Mouth Abnormalities/
 593 86 hare lip\$.tw.
 594 87 harelip\$.tw.
 595 88 Palatoschisis.tw.
 596 89 cleft lip\$.tw.
 597 90 cleft palate\$.tw.
 598 91 orofacial\$ cleft\$.tw.
 599 92 facial cleft\$.tw.
 600 93 oral cleft\$.tw.
 601 94 craniofacial cleft\$.tw.
 602 95 or/22-94
 603 96 21 and 95
 604 97 exp animals/ not humans/
 605 98 96 not 97
 606 99 limit 98 to "all adult (19 plus years)"
 607 100 limit 98 to "all child (0 to 18 years)"
 608 101 100 not 99
 609 102 98 not 101
 610

611 Table 1: Risk of bias in RCTs

Study	Study Design	Sequence Generation	Allocation Concealment	Method of blinding of outcome assessor	Completeness of outcome data	Reporting of outcome data
Bessell et al (2012)	RCT	Low	Low	Low	Low	Low
Srivastava & Chaudhury (2014)	RCT	Unclear	Unclear	Unclear	Low	Low

RCT = Randomised controlled trial, Low = low risk of bias, High = high risk of bias, unclear = information in the paper not sufficient to assess risk of bias

614 Table 2: Risk of bias observational studies

Study	Study Design	Randomisation Procedure	Attrition	Measurement
Semple et al (2009)	Observational	High	Low	Low
Jolly et al (2010)	Observational	Unclear	Unclear	Unclear

616 Low = low risk of bias, High = high risk of bias, unclear = information in the paper not sufficient to assess risk of bias

Table 3: Characteristics of included studies

Study	N	Location	Population	Age	Study Design	Intervention	Comparator intervention	Setting	Facilitator	Intensity	Duration	Follow-up
Srivastava & Chaudhury (2014)	90*	India	Adults with amputations	22-52 yrs	RCT	Person-centred counselling	Treatment as usual	Not stated	Psychiatric nurse	6 weekly sessions	Not stated	No follow up reported
Bessell et al 2012	83 (49 f)	United Kingdom	Adults with any visible difference	18+	RCT	CBT/SST	No treatment control	Clinic	Therapist/self help	8 weekly sessions	1 hour	6 month post-intervention
Jolly et al (2010)	15 (15 f)	United States	Women with Lupus	18+	CT	CBT/cosmetic training	No treatment control	Clinic	Therapist	10 weekly sessions	1.75 hours	Week 18 & 24 post intervention
Semple et al 2009	54 (28 F)	United Kingdom	Head and neck cancer patients	31-75 yrs	CT	CBT/SST	Usual care	home	Clinical nurse specialist	2-6 fortnightly sessions	90 mins	3-month follow-up

*Not all studies reported gender. Figures are provided where reported

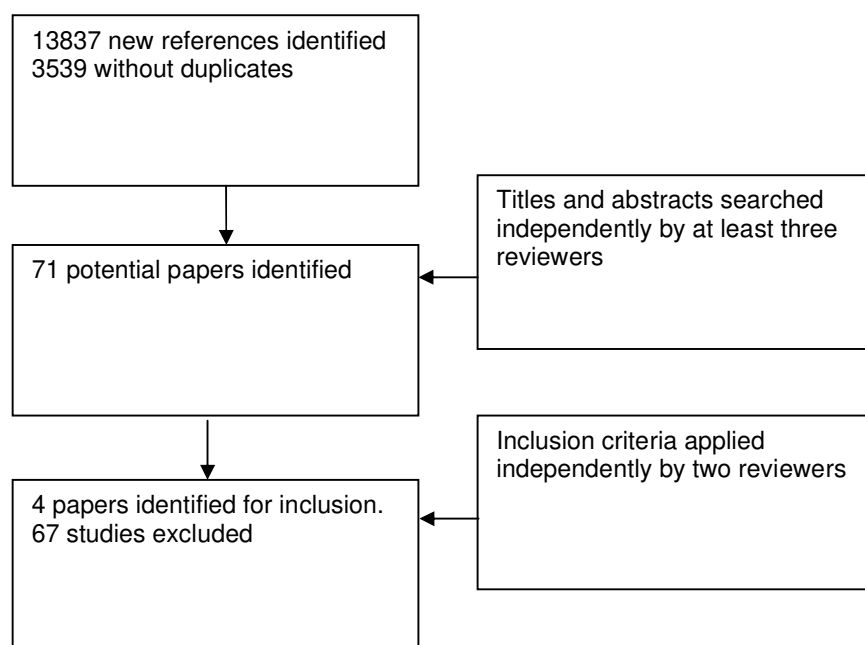


Figure 1: Flow diagram of search results