

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

2 Running title: Adults and visible difference review

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## 15 Abstract

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

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

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

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

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

33 Keywords:

- 34 • Visible differences, Psychosocial, narrative synthesis, Cognitive-behavioural therapy,  
35 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 12<sup>th</sup> 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.

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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 review<sup>1</sup>. Secondary measures included measure of  
151 anxiety and depression and general improvements in psychological symptoms, interpersonal

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<sup>1</sup> 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.

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

154

155 Exclusion Criteria

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

164

165 Ethical considerations

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

168

169 Analysis

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

174



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

## 199 Results & Discussion

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

205 Insert figure 1 here

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

### 215 Risk of Bias Assessment

216 Two papers (Srivastava & Chaudhury, 2014; Bessell et al, 2012); were assessed using the  
217 Cochrane risk of bias assessment tool which is suitable for assessing RCTs (Higgins & Green,  
218 2011). The Bessell et al (2012) paper was assessed for risk of bias by two researchers  
219 independent of the paper's authors (AM & JG) as two of the authors were also the authors  
220 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, Kernohen &  
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.

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

300

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

305 Effects of Interventions: Timing of Intervention

306 This review attempted to identify the optimal duration and intensity of intervention. The  
307 studies included within this review varied in duration from two sessions (Semple et al,  
308 2009), through to 10 sessions (Jolly et al, 2010). Full details of intervention duration can be  
309 found in Table 3. The intensity of the interventions consisted of weekly (Srivastava &  
310 Chaudhury, 2014; Bessell et al, 2012) or fortnightly sessions (Semple et al, 2009). Sessions  
311 were between one and two hours in length (see Table 3 for full details of intensity).

312 Due to the differing intensity and duration across the studies, it is difficult to draw any firm  
313 conclusions regarding the optimal length and intensity of therapy. However, most studies  
314 opted for between 6 - 10 sessions administered weekly for 1-1.5 hours. Therefore, it would  
315 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

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

395

396 Implications for future research, policy and practice

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

409 The current interventions have provided very limited support for the CBT and combined  
410 CBT and SST models. These techniques offer individuals practical solutions to some of their  
411 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

436 Acknowledgements

437 Thanks to Julie Griffin, Natasha-Ann Deeprise, Hannah Wadland, Amir Massoumian and Jo-  
438 Lee Hankinson for support with title and abstract checking and risk of bias assessment.

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? (2<sup>nd</sup> 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.ti.  
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 tumo?r\$)).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

612 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

613

614 Table 2: Risk of bias observational studies

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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

617 Table 3: Characteristics of included studies

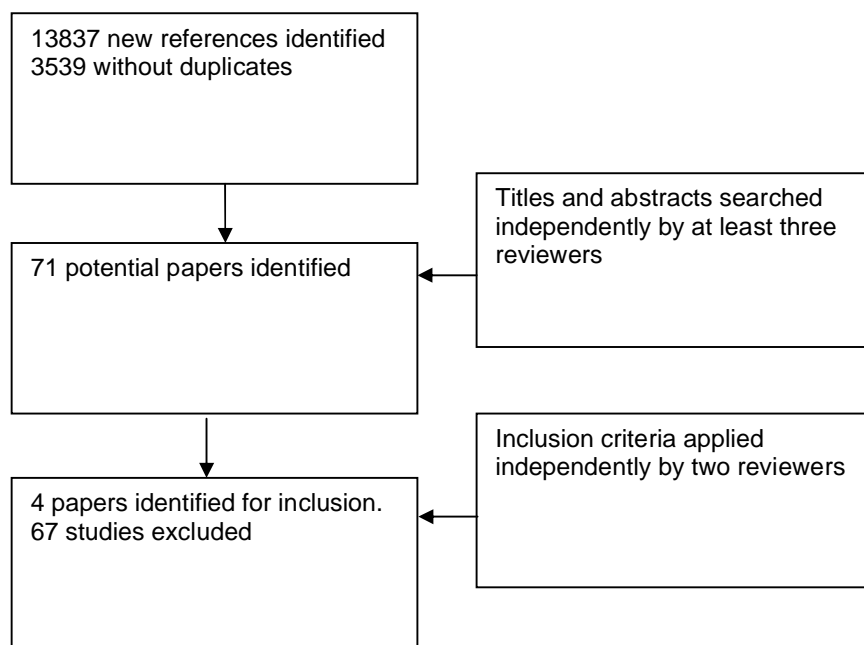
618

Study	N	Location	Population	Age	Study Design	Intervention	Comparator intervention	Setting	Facilitator	Intensity	Duration	Follow-up
<b>Srivastava &amp; Chaudhury (2014)</b>	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
<b>Bessell et al (2012)</b>	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
<b>Jolly et al (2010)</b>	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
<b>Semple et al (2009)</b>	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

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

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**Figure 1: Flow diagram of search results**