Factors affecting subjective appearance evaluations among patients with congenital craniofacial conditions: An application of Cash’s cognitive-behavioural model of body image development

Abstract

Satisfaction with appearance is of central importance for psychological well-being and health. For individuals with an unusual appearance, such as congenital craniofacial anomalies (CFA), appearance evaluations could be especially important. However, few, if any papers have presented a comprehensive synthesis of the factors found to affect subjective satisfaction with appearance among children, adolescents, and adults born with a CFA. Further, only a handful of craniofacial studies have applied psychological theories or models to their findings, resulting in an overall lack of guidance for researchers in the field. This paper summarises the literature pertaining to satisfaction with appearance among those affected by CFAs, and examines the extent to which Cash’s cognitive-behavioural model of body image development (2012) fits with this literature. Given the overlap between factors of interest in the field of CFAs, and in the area of body image more broadly, a closer collaboration between the two research fields is suggested.

Key words: Subjective, appearance, satisfaction, cognitive-behavioural model, craniofacial, cleft.
Introduction

Satisfaction with appearance is of central importance for individuals’ psychological well-being and health (Cash & Pruzinsky, 2002; Rumsey & Harcourt, 2004). Consequently, psychological research has investigated a range of factors thought to affect the development of self-perceptions of appearance. In addition to empirical research, theoretical approaches have also been described when exploring the development of subjective appearance evaluations. One comprehensive attempt to capture the broad range of factors and processes potentially involved in the development of subjective satisfaction with appearance is Cash’s cognitive-behavioural model of body image development (2012; see Figure 1). The model includes (a) predisposing and social learning experiences that affect how people think, feel, and act in relation to their body, and (b) current or proximal factors that precipitate or maintain subjective evaluations of body image. Predisposing factors consist of cultural socialisation, interpersonal experiences, physical characteristics, and personality attributes. Proximal factors consist of appearance-schematic processing, activating events, internal dialogues, body image emotions, and self-regulatory actions. Additionally, satisfaction with body image is influenced by appearance investment and evaluations. Cash’s cognitive-behavioural model is a guide to understanding satisfaction with appearance from a multidimensional perspective, with an additional emphasis on the complex interactions between external events and internal factors (Cash, 2012).

The centrality of subjective appearance evaluations is illustrated by the range of emotional, social, and behavioural factors that have been found to be associated with appearance, both within the general population (Cash & Pruzinsky, 2002; Rumsey & Harcourt, 2012) and among clinical samples (Feragen & Stock, 2017; Hunt, Burden, Hepper, & Johnston, 2005; Muftin & Thompson, 2013; Rumsey, 2012; Stock & Feragen, 2016; Thompson & Kent, 2001).
One group thought to be particularly vulnerable to dissatisfaction with appearance is individuals who are affected by a visible difference, such as a congenital craniofacial anomaly (CFA). CFA is a broad term used to describe a wide range of conditions affecting the head and neck (Holmbeck & Aspinall, 2015), with prevalence rates and characteristics varying widely across conditions (Buchanan, Xue, & Hollier, 2014). This includes the most common CFA, cleft lip and/or palate (CL/P), in addition to rarer conditions, such as craniosynostoses and Treacher Collins syndrome, among many others. In spite of surgical and other interventional procedures from birth through to adulthood, it is unlikely that the visible difference will be removed entirely, and people with a CFA may therefore feel that they differ from their peers in terms of facial appearance. Many studies have explored professional and lay evaluations of patients’ appearance and treatment outcomes (e.g. Collett et al., 2013; Meyer-Marcotty, Gerdts, Stellzig-Eisenhauer, & Alpers, 2011; Meyer-Marcotty & Stellzig-Eisenhauer 2009), while others have focused on patients’ subjective perceptions (e.g. Plomp, Versnel, van Lieshout, Poublon, & Mathijssen, 2013; van Lierde et al., 2012; Wehby et al., 2012).

Research within the CFA field has also investigated associations between satisfaction with appearance and other psychological variables, such as overall quality of life (Crerand, Sarwer, Kazak, Clarke, & Rumsey, 2017; Mani, Reiser, Andlin-Sobocki, Skoog, & Holmström, 2013; Marcusson, Paulin, & Ostrup, 2002; Oosterkamp et al., 2007; Roberts & Mathias, 2013), in addition to social, emotional, and psychosocial adjustment (Bilboul, Pope, & Snyder, 2006; Marcusson et al., 2002; Roberts & Mathias, 2013). Nonetheless, knowledge about specific factors thought to affect the development of subjective appearance evaluations among those with CFAs is sparse, and the lack of longitudinal studies complicates our full understanding of the development of self-evaluations of appearance. Further, the application of appropriate models and theories is significantly lacking within craniofacial research (Baker, Owens, Stern, & Willmot, 2009; Stock, Feragen, Moss, & Rumsey, in press). Few, if any recent papers, have presented a comprehensive synthesis of the factors found to affect subjective satisfaction with appearance among children, adolescents, and adults born with a CFA, or
considered the degree to which the craniofacial literature addresses similar issues to those reported within the general population.

This article summarises the available literature pertaining to satisfaction with appearance among those affected by CFAs, in order to explore factors that could explain the development of, and variation in subjective appearance evaluations. In 2005, Hunt and colleagues reviewed the literature examining psychological adjustment to CL/P between 1982 and 2003, while a more recent review systematically identified studies published in the field of CL/P from 2004 until 2015 (Stock & Feragen, 2016). One further review summarised findings related to psychological adjustment to other rare CFAs between 2000 and 2016 (Feragen & Stock, 2017). Papers investigating satisfaction with appearance from all three of these reviews were identified, and included in the present work. An updated search was also performed in June 2017 and xx papers were added. Across all three reviews and in the updated search, databases were examined using diagnostic search terms (Cleft - Cleft lip and palate - Cleft lip/palate - CLP - CL/P - Craniofac* - Visible difference - Disfigure*- Impair* - Treacher Collins - Crouzon - Apert - Craniosynostos* - Microsomia - Muenke - Sæthre-Chotzen - Pfeiffer - Goldenhar - Trigonocephaly), in addition to appearance-related search terms (Appearance - Satisfaction - Surg* - Function - Rating - Outcome - Body Image). The Boolean operators OR were used between search terms within diagnostic terms and appearance-related search terms, while the operator AND was used between the two categories. Studies exploring satisfaction with appearance without addressing possible associations with other variables were not systematically included in the present work, since overviews of this research can already be found in the literature.

An overview of all included papers can be found in Table 1. Drawing upon additional literature from the broader field of visible difference for contextual purposes, this paper also examines the extent to which current understanding of appearance satisfaction among individuals born with CFAs aligns with Cash’s cognitive-behavioural model of body image development (2012), with the ultimate aim of guiding future research within both fields.
Predisposing “historical” and developmental factors

Historical and developmental variables are described as predisposing individuals to the acquisition of certain subjective appearance attitudes (Cash, 2012). The cognitive-behavioural model (Cash, 2012) includes four main factors that are thought to influence how people come to think, feel, and act in relation to their appearance: cultural socialisation, interpersonal experiences, physical characteristics, and personality attributes.

Cultural socialisation

Standards of beauty and attractiveness are rooted in sociocultural norms, and convey information about society’s and individuals’ attitudes towards appearance. Almost half a century ago, Goffman (1968) used the term ‘stigma’ to refer to the relationship between a person’s characteristics and the value society places on appearance. Since then, a wide range of literature has investigated societal acceptance of facial differences and other visible disabilities, identifying underlying negative assumptions, face-value judgements, and instances of prejudice (Bull & Rumsey, 1988; Chan, McPherson, & Whitehill, 2006; Goode, Ellis, Coutinho, & Partridge, 2008; Macgregor, 1990; Stock, Whale, Jenkinson, Rumsey, & Fox, 2013). It has been observed that societal attitudes to visible difference have improved to some degree in recent years (Pausch et al., 2016). Nevertheless, social discrimination on the basis of an unusual appearance is still described in the literature (Partridge, 2010), and research has demonstrated an association between perceived social discrimination and reduced satisfaction with appearance (Roberts, 2014). Further, adults with a visible difference have described the ongoing challenge of living in a modern society that simultaneously tries to accept people who look different while placing more emphasis on appearance than ever before (Hamlet & Harcourt, 2015).

While several studies have examined societal attitudes to visible difference, few have examined how these perceptions are directly or indirectly perceived, or how they influence subjective satisfaction with appearance among those affected by a CFA. Only one quantitative study was identified, which
revealed a moderate association between satisfaction with appearance and perceptions of discriminations in adults with CFA (Roberts, 2014). Other studies outline clinical case-studies and qualitative interviews with people with personal experience in living with a CFA (Cadogan & Bennun, 2011; Searle, Neville, & Waylen, 2017; Stock, Feragen, & Rumsey, 2016; Tevik & Feragen, 2015). While these studies have voiced young people’s and adults’ concerns in relation to their interaction with sociocultural attitudes and the pressure created through media ideals, they have not specifically addressed the subsequent impact of this on subjective appearance evaluations. Several studies seem to assume that societal attitudes are negative, and equally assume a negative psychosocial impact of such attitudes on people living with a visible difference, without testing whether this is actually the case. A recent study investigating implicit attitudes towards people with a visible difference provide preliminary evidence that attitudes are not necessarily negative, and should instead be explained by people’s surprise at seeing something unusual, and uncertainty regarding how to behave (Roberts, Neate, & Gierasch, 2017). Taken together, these findings represent a possible gap in the craniofacial literature, and thus a weak link when applying this work in the context of Cash’s model (2012). Another limitation in the general literature, as well as in the craniofacial literature is that most research is performed within Western cultures. Studies addressing societal perceptions of individuals with CFAs in non-Western countries have shed light on social and structural inequalities due to misconceptions about congenital medical conditions, which could potentially impact on body image and should also be addressed by future research (Adeyemo, James, & Butali, 2016).

**Interpersonal experiences**

Appearance-related socialisation involves more than cultural messages and media-based messages. Expectations, opinions, comparisons, and criticisms are communicated through verbal and non-verbal interactions with people who may express, model, and reinforce cultural expectations, including family members, friends, and strangers (Cash, 2012). More specifically, appearance-related teasing in the family, and especially from siblings, has been shown to be associated with body
dissatisfaction (Berge, Hanson-Bradley, Tate, & Neumark-Sztainer, 2016; Keery, Boutelle, van der Berg, & Thompson, 2005). In addition, positive and negative social interactions could impact on and contribute to the formation of an individual’s appearance evaluations (Cash, 2012).

Associations between social experiences and subjective appearance evaluations have been described in a number of cross-sectional and qualitative studies within the CL/P literature (Berger & Dalton, 2011; Feragen & Borge, 2010; Feragen, Borge, & Rumsey, 2009; Feragen, Kvalem, Rumsey, & Borge, 2010), as well as in qualitative research (Beaune, Forrest, & Keith, 2004; Cadogan & Bennun, 2011; Hamlet & Harcourt, 2015; Havstam, Laakso, & Ringsberg, 2011; Searle et al., 2017; Stavropoulos, Hallberg, Mohlin, & Hagberg, 2011; Stock et al., 2016). Associations between subjective appearance evaluations and social experiences have been shown to be stronger among adolescents (Feragen et al., 2010) than in younger children (Feragen & Stock, 2016). Another study examined a positive correlation between the self-perceived influence of social and professional life and subjective assessments of appearance (Gkantidis, Papamanou, Christou, & Topouzelis, 2013). One further study on adolescents with CL/P (Feragen, Stock, & Kvalem, 2016) identified an association between self-perceptions of romantic appeal and satisfaction with appearance. Other research has referred to the visibility of the craniofacial condition as a direct cause for other people’s reactions or teasing (e.g. Searle et al., 2017), referring to frameworks such as Goffman’s stigma theory (1968), without necessarily discussing the role of internal processes, attributions, and interpretations in the individual’s perceptions of interpersonal experiences.

People’s attention is automatically drawn to aspects of appearance that differ from what is expected. As a consequence, a craniofacial condition may trigger overt or subtle social reactions, such as staring, whispering, comments, and/or questions (Feragen, 2012). Several studies have shed light on individuals’ self-consciousness and distress when meeting new people, or when entering unfamiliar social environments (Cadogan & Bennun, 2011; Stock et al., 2016; Turner, Thomas, Dowell, Rumsey, & Sandy, 1997). People affected by a CFA will inevitably be confronted by other people’s reactions to
the visible difference, and over time will hopefully learn to cope (Havstam et al., 2011). Yet, individuals with CFAs have also described how ambiguous, judgmental, and/or negative interactions can become incorporated into their sense of self-worth (Beaune et al., 2004; Stavropoulos et al., 2011). In the general population, negative interpersonal experiences, such as appearance-related criticism, have been shown to predispose individuals to appearance dissatisfaction (Cash, 2012). While one study did not support this assumption within the CFA literature (Lorot-Marchand et al., 2015), other studies have linked perceived peer harassment and teasing to lower satisfaction with appearance among young people with CFAs (Feragen & Borge, 2010; Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Searle et al., 2017). In addition, a recent longitudinal study demonstrated the detrimental long-term effect of self-perceived teasing on subjective satisfaction with appearance, most notably among adolescent females (Feragen & Stock, 2016).

More recently, the relationship between positive social interactions and satisfaction with appearance has also been addressed in the craniofacial literature. For example, associations have been demonstrated between social acceptance, close friendships, and subjective satisfaction with appearance among adolescents with CL/P (Feragen et al., 2010). A positive association has also been found between satisfaction with appearance and peer relationship quality (Shapiro, Wajee, Ranganathan, Buchman, & Warschausky, 2015), and with perceived peer and parent support (Shute, McCarthy, & Roberts, 2007) in samples of young people with a range of CFAs.

In summary, the craniofacial literature seems to have addressed possible associations between interpersonal experiences and appearance satisfaction, ranging from subtle social reactions to overt teasing and bullying, and has explored the impact of supportive, as well as detrimental social experiences. Surprisingly, and in light of Cash’s model, little research has directly addressed the impact of social experiences on the development of subjective appearance evaluations. Further, many studies base their analyses on the assumption of a direct link between the visible difference and appearance dissatisfaction, hence obscuring our understanding of the associations between interaction experiences.
and appearance. Last, no identified studies investigated appearance-related talk, appearance attitudes, or family dynamics in relation to CFAs. For example, future studies could address discrepancies in the motivators for surgery between parents and their children, and its impact on the child’s appearance satisfaction. Little is known about whether children with craniofacial conditions are teased by family members, and how this could impact on the development of body image.

**Physical characteristics**

According to Cash (2012), body image development is affected by one’s physical characteristics, and these characteristics impact how people are perceived and treated by others. Social feedback and internal cognitive self-appraisals are thought to mediate this relationship (Cash, 2012). The cognitive-behavioural model also includes the potential impact of a visible difference in relation to the individual’s appraisal of subjective appearance satisfaction, and how well the person feels his or her appearance matches social standards (Cash, 2012).

Several studies have suggested that the “objective” visibility caused by the craniofacial condition may lead to reduced satisfaction with appearance when compared to controls (Crerand et al., 2016; Oosterkamp et al., 2007; Slifer et al., 2004; Versnel, Duivenvoorden, Passchier, & Mathijssen, 2010). In addition, adolescents with a visible cleft (cleft lip; cleft lip and palate) were found to be less satisfied with appearance than those with a non-visible cleft (palatal involvement only) in a number of other studies (Berger & Dalton, 2009; Feragen & Stock, 2016a; Feragen, Stock, Sharratt, & Kvalem, 2016), and to be less satisfied with facial features affected by the craniofacial diagnosis than other aspects of appearance (Feragen et al., 2015). In contrast, other studies have shown no associations between the “objective” visibility of the condition and subjective appearance satisfaction (e.g. Feragen & Stock, 2016a), and have instead underlined the importance of distinguishing ‘objective’ (visible vs non-visible) from subjective evaluations of appearance. In the wider field of visible difference, research has clearly shown the objective visibility of the condition to be a weak predictor of the patient’s adjustment to the challenges of
living with a visible difference (Appearance Research Collaboration, 2009; Feragen, 2012; Moss, 2005; Rumsey & Harcourt, 2004). Rather, it is the individual’s subjective satisfaction, and the extent to which he or she believes the condition is visible to others (Stock et al., 2016), that is the best predictor of psychological adjustment, well-being, and quality of life. This central aspect of living with a visible craniofacial condition could be in line with Cash’s cognitive-behavioural model (2012), in which social feedback and internal cognitive self-appraisals of subjective appearance satisfaction are thought to mediate and interact with physical characteristics (Cash, 2012). The lack of longitudinal studies in the craniofacial field impede our understanding of the shifting nature of subjective evaluations, not only as related to social interaction experiences, but also as a function of age and life experience; perspectives that should be addressed by future research.

**Personality attributes**

Individual personality attributes have been shown to influence the formation of appearance attitudes, with self-esteem being described as the most pivotal of these factors (Cash, 2012). Attachment style, attitudes and values, perfectionism, public self-consciousness, and a need for social approval are all described as personal attributes that may impact on subjective appearance evaluations (Cash, 2012).

In the 1980s, some early research on CL/P examined associations between personality attributes and adjustment problems (Richman, Holmes, & Eliason, 1985), based on the assumption that concerns with facial appearance could lead to social introversion (Richman, 1983). More recent research seems to categorise personality attributes as one potential factor that partly explains individual variations in adjustment, rather than as a direct consequence of living with a visible difference (Appearance Research Collaboration, 2009).

Within the CFA literature, self-esteem has been linked to appearance satisfaction among adults with a congenital CFA in two papers (Stock et al., 2016; Versnel et al., 2010), and presented as a factor which is more important to subjective appearance evaluations than demographic factors or physical
characteristics (Versnel et al., 2010). Self-worth has also been shown to be highly associated with satisfaction with appearance among adolescents with CFAs (Bilboul et al., 2006).

Although attachment style has been discussed within the CFA literature, alongside the assumption that the infant’s visible difference may interfere with the attachment process, conflicting results have been reported, and direct associations between attachment style and subjective satisfaction with appearance have not been investigated (Despars et al., 2011; Gassling et al., 2014; Montiroso et al., 2012; Murray et al., 2008). Dispositional style and other personality attributes, such as self-worth, toughness, and inner strength have also been explored, predominantly by qualitative studies (Cadogan & Bennun, 2011; Eiserman, 2001; Havstam et al., 2011; Stock et al., 2016), or in the context of visible difference more generally (Appearance Research Collaboration, 2009). Although public self-consciousness and a need for social approval have not been investigated directly among those born with CFAs, the related concept of ‘fear of negative evaluation’ has received some attention. For example, one study reported a strong correlation between the fear of negative evaluation and satisfaction with appearance in adults with CFAs (Roberts & Mathias, 2013), while another study of adolescents reported a moderate to strong association (Shute et al., 2007). Similarly, adults with CFAs were more satisfied with their facial appearance if they reported low fear of negative evaluation, which again was found to be substantially more important than demographic or physical determinants (Versnel et al., 2010).

Quantitative studies investigating dispositional style and personality attributes and their associations or impact on subjective appearance evaluations are scarce within the CFA literature. Qualitative research, on the other hand, has more systematically explored personality attributes that may strengthen or decrease an individual’s ability to cope with the challenges of living with CFA. However, while qualitative research contributes important knowledge about internal processes and individual variations, findings need to be investigated quantitatively in larger and representative samples in order for conclusions to be generalised to the population under study. Findings pointing to protective dispositional factors (Cadogan & Bennun, 2011; Eiserman, 2001; Havstam et al., 2011; Stock et al.,
 Appearance attitudes and schemas

Appearance attitudes, or dispositional appearance evaluations, are thought to function as central organising constructs, acquired through the interplay and constant interaction between developmental and environmental factors (Cash, 2012), such as those described in the previous section of this paper. Two basic attitudinal elements are included in Cash’s model: appearance investments and evaluations. In addition, a higher-order attitudinal construct included in the model, acting as a central body image construct, is that of self-schemas. Appearance-schematic processes are described as cognitive generalisations about the self, which are derived from past experiences that guide the organisation of current social experiences and processes (Markus, 1977). The appearance self-schema represents both the emotional evaluations of the self (valence) and its organising structure (Moss & Rosser, 2012). Self-schemas therefore influence our thoughts, behaviours, and emotions, and reflect one’s core, affect-laden assumptions or beliefs about the influence and importance of appearance in life, including the centrality of appearance to one’s sense of self (Cash, 2012).

 Appearance investments

Appearance investment is described as the cognitive-behavioural importance people place in their appearance (Cash, 2012). In the craniofacial literature, only one quantitative study has explored the relationship between appearance investment and appearance satisfaction, in which adolescents with CFAs reported lower levels of appearance investment than controls, while not differing from the control group in relation to appearance satisfaction, and with xx being unrelated to overall quality of life (Crerand et al., 2017). In a qualitative study with older adults with CL/P, the importance of managing appearance in order to reduce unwanted social reactions to their visible difference was presented (Hamlet
& Harcourt, 2014). Yet, another qualitative study drew inferences in relation to a possible negative association between high appearance valence and appearance dissatisfaction (Stock et al., 2016).

As discussed in Crerand et al. (2017), differences in appearance investment could be interpreted in several ways, depending on the sample under study. In some samples, high levels of investment may indicate or predict body dissatisfaction. In contrast, in samples of adolescents with a visible condition, low levels of investment could be seen as an adaptive or protective behaviour, reflecting efforts to build up other aspects of the self-concept by minimising the importance of appearance (Crerand et al., 2017). More research is needed in order to better understand the role of appearance investment among people living with a visible difference. Given the central aspect of appearance in CFAs, surprisingly little research has investigated the construct of appearance investment and its importance. Again, qualitative studies are the main contributing sources of knowledge, and more systematic and generalisable investigations are therefore needed within the CFA literature.

**Appearance evaluations**

Appearance evaluations refer to the positive-to-negative appraisals of and beliefs about one’s appearance (Cash, 2012), such as satisfaction or dissatisfaction with appearance. According to the model, appearance evaluations stem from the degree of discrepancy or congruence between self-perceived appearance characteristics and subjective appearance ideals (Cash, 2012).

An extensive body of literature has explored satisfaction and dissatisfaction with appearance in the context of CFAs (see Feragen & Stock, 2017; Hunt et al., 2005; Stock & Feragen, 2016). Dissatisfaction with appearance, and specifically with the facial features affected by the condition, seems to be a prevalent concern among those with CL/P (e.g. Berger & Dalton, 2009; Hunt et al., 2006; Semb et al., 2005). However, craniofacial studies involving comparison or control groups have concluded that levels of dissatisfaction with appearance among those with CFAs may in fact be comparable to that seen in the general population (see Feragen & Stock, 2017; Stock & Feragen, 2016). The diversity of findings
in the craniofacial literature therefore calls for researchers to move away from the paradigm that a visible difference will automatically lead to dissatisfaction with appearance, and toward an exploration of the factors which may influence an individual’s self-perceptions, as sought in the present paper.

Appearance evaluations and self-schemas are seen as central elements of appearance attitudes in the cognitive-behavioural model (Cash, 2012). The measurement of appearance valence and salience has been validated in a large sample of adults with a visible difference (congenital and acquired conditions), in addition to their independent and combined role in the processing of appearance-related information (Moss, Lawson, & White, 2014). Valence is defined as the emotional evaluation of the self in relation to appearance, whereas salience refers to the centrality and accessibility of appearance-related information (Moss et al., 2012). The appearance self-schema has been presented as the aspect of the self-concept which represents both this emotional evaluation (valence), as well as its organising structure (salience), which have been linked to the interpretation and perception of psychosocial experiences reported by people with appearance concerns (Moss et al., 2012). Almost twenty years ago, Thompson and Kent (2001) addressed the need for future research within the field of visible difference to investigate concepts such as self-schemas. Unfortunately, the CFA literature still fails to systematically examine the role of these concepts in explaining individual variations in appearance satisfaction. The investigation of cognitive generalisations about the self, their role in the perception and interpretation of social experiences, and their influence on thoughts, behaviours and emotions, could be an interesting area of future research, and could contribute to a better understanding of the influence and importance of appearance among individuals living with a visible difference.

**Proximal events and processes**

According to the cognitive-behavioural model (Cash, 2012), situational cues or contextual events will potentially activate schema-driven processing of information. Current life events consist of precipitating and maintaining influences on appearance experiences (Cash, 2012). Factors that are
described in the model are: activating events, internal dialogues, body image emotions, and self-regulatory strategies and behaviours.

**Activating events**

According to Cash’s model, situational cues or specific events may activate schema-driven processing of information about, and self-evaluations of, one’s appearance. Markus (1977) suggests that schema-related information is more easily recalled from memory, whereas counter-schematic information is distorted or ignored. As a consequence, appearance schemas contribute to cognitive bias (Kent, 2000), giving more importance and attention to information that confirms the individual’s appearance-related self-schema. The more central an individual perceives appearance to be, the more salient and accessible information will be that triggers appearance-related self-worth. Hence, if valued aspects of the self are perceived as flawed, this will have a greater impact on global self-worth (Moss et al., 2012). Specific psychosocial experiences and events may trigger self-schemas, and confirm the individual’s expectations of social interactions. Examples of precipitating events include appearance or mirror exposure, social situations involving appearance evaluations, and changes in appearance (Cash, 2012). Although no studies have investigated appearance or mirror exposure systematically in relation to CFAs, both negative social interaction experiences, and experiences triggered by appearance-altering treatment could be categorised as activating events.

Looking ‘different’ may evoke appearance-related teasing from others, as well as staring, comments and questions (which may also be interpreted by the individual as ‘teasing’). The frequency of self-reported teasing among those affected by CFAs is extremely variable between studies (e.g. Hunt et al., 2006; Lorot-Marchand et al., 2012; Semb et al., 2005). In the context of this paper, few such studies have explicitly and directly measured the impact of teasing on subjective appearance satisfaction, except for one recent study that demonstrated the detrimental long-term effect of teasing on dissatisfaction with appearance among adolescents with a craniofacial condition (Feragen & Stock, 2016). Future longitudinal studies are needed if we are to disentangle the complex interplay between
negative social events and subjective appearance evaluations. Unfortunately, these are lacking within the craniofacial literature (Stock & Feragen, in press). Investigations of a romantic break-up as a potential activating event is currently also missing from the CFA literature, although one study did discuss the potential impact of romantic experiences on appearance evaluations (Feragen et al., 2016).

In spite of the complex and long-term nature of the care pathway for many craniofacial conditions, few studies have specifically examined patients’ experiences of undergoing appearance-altering treatment and its impact upon appearance satisfaction (Feragen & Stock, 2017). Through the use of qualitative methodology, however, young people and adults with CFAs have described the physical and psychological challenges related to undergoing appearance-altering treatment, and how these changes can trigger an emotional crisis, and a need to revisit their appearance-related identity (Beaune et al., 2004; Stavropoulos et al., 2011; Stock, Feragen, & Rumsey, 2015). As an illustration, adults have reported that seeing their reflections in the mirror for the first time after an operation can trigger strong emotional reactions (Cadogan & Bennun, 2011). In contrast, the gradual ‘normalisation’ of appearance via surgical interventions have also been linked to a perceived reduction in social visibility and improved psychological adjustment among adults with CFAs (Cadogan & Bennun, 2011; Stavropoulos et al., 2011). A number of further quantitative studies have reported improved perceptions of appearance as a result of surgical or orthodontic treatment among patients with CFAs (Byrne, Chan, & O’Broin, 2014; Hens et al., 2011; Landsberger et al., 2006). Patients may also choose to decline appearance-altering surgery due to satisfaction with appearance (Searle et al., 2017), or choose appearance-altering surgery as a way to improve self-perceptions of appearance (Stock et al., 2016), with corresponding changes in social visibility and self-perceptions (Tevik & Feragen, 2015). However, and in contrast, an equal number of studies have suggested that undergoing more surgical interventions and/or reporting more desire for corrective surgeries may be linked to higher appearance dissatisfaction (Mani et al., 2013; Nkenke, Stelzle, Vairaktaris, & Knipfer, 2013; Stock et al., 2015), where the aesthetic outcomes of
treatment do not always outweigh the emotional and physical investment (Bemmels et al., 2013; Stock et al., 2015).

**Internal dialogues (thoughts, interpretations, and conclusions)**

According to the cognitive-behavioural model (Cash, 2012), internal dialogues involve emotion-laden automatic thoughts, inferences, interpretations, and conclusions about one’s appearance. Internal appearance dialogues may include distorted perceptions of events or attitudes, dichotomous thinking, emotional reasoning, social comparisons, skewed inferences, overgeneralisations, and a magnification of appearance-related problems (Cash, 2012). Internal dialogues may be challenging to examine with a quantitative methodology, and few studies seem to investigate these concepts in relation to appearance. Qualitative methodology, on the other hand, has been used more frequently within the CFA literature in relation to internal dialogues, pointing to the varied and individual aspects of processing and interpreting social interactions.

In one such qualitative study, some adults affected by CL/P reported that having a visible difference had made them more or less susceptible to societal beauty ‘ideals’, and more or less concerned with their appearance (Stock et al., 2015). Another qualitative study of CFAs also described the presence of inner strength and self-confidence as potential protective factors in situations where satisfaction with appearance was challenged (Eiserman, 2001). In a third qualitative study, with adults born with Treacher Collins syndrome, it was described how cognitively reconstructing social and emotional experiences had helped adults to become more socially assertive, to utilise positive coping strategies as a buffer for negative experiences, and to become more self-accepting (Beaune et al., 2004). Re-framing and re-categorisations (Searle et al., 2017), as well as variations in individual perceptions of social interpersonal experiences (Stock et al., 2016) have also been discussed in relation to adults with CL/P. Adults with Crouzon syndrome have highlighted the importance of “making the best of your situation” (Stavropoulos et al., 2011). On the other hand, the opposite may be the case. Stares and
questions may be interpreted negatively by children who are emotionally vulnerable, even if such behaviors originally stemmed from neutral curiosity (Feragen et al., 2009).

In order to enhance our understanding of the development of self-evaluations of appearance, we need research that includes or controls for individual differences in the internal processing of appearance-related information. Well-known discrepancies between subjective self-reports and information provided by other informants (De Los Reyes & Ohannessian, 2016) illustrates the uniqueness of each individual’s thoughts and interpretations when processing our lived experiences. Collecting the perspectives of multiple informants in order to triangulate data from different sources is important, however without underestimating the centrality of self-perceptions for psychological health and function. Future research, investigating internal appearance dialogues, such as distorted perceptions and interpretations of social interactions, social comparisons, overgeneralisations, and a magnification of appearance-related problems (Cash, 2012) would shed light on the process of individual adjustment to the challenges associated with a CFA.

**Body image emotions**

Appearance-related emotions are described in Cash’ cognitive-behavioural model (2012) as affectively-toned evaluations and investments that are the result of activating events and cognitive processing. Body emotions may be positive (e.g., appreciation or pride) or negative (e.g. shame or anxiety). Certain aspects of emotional wellbeing among individuals born with CFAs may be relevant, since emotional health and satisfaction with appearance are strongly related in the craniofacial literature (see Feragen and Stock, 2017; Hunt et al., 2005; Stock & Feragen, 2016). For example, one study identified an association between subjective satisfaction with appearance and emotional adjustment in adults with CL/P (Ramstad, Ottem, & Shaw, 1995). Similar associations were also identified in adolescents with CL/P at age 16 (Feragen & Stock, 2015); a correlation that was not found to be significant at the earlier age of 10 (Feragen et al., 2016). Another study of children with CL/P also found
an association between emotional adjustment and satisfaction with appearance (Wehby, 2012). Equally, dissatisfaction with facial appearance was found to be significantly correlated with depression and emotional distress in adults with a variety of CFAs (Marsusson et al., 2002; Roberts & Mathias, 2013), particularly among females (Feragen et al., 2015; Shapiro et al., 2015). In summary, the association between emotional distress and dissatisfaction with appearance has been clearly addressed by the craniofacial literature. However, all referred studies are based on cross-sectional samples that do not allow for an interpretation of directionality. Longitudinal analyses based on representative and large samples should therefore be a priority for future craniofacial research.

**Adjustive self-regulatory strategies and behaviours**

In order to cope with distressing appearance thoughts and emotions, individuals develop and engage in a range of actions and reactions aimed at promoting adjustment to the challenges of activating events (Cash, 2012). Adjustive strategies may involve avoidance or concealment behaviours, seeking social reassurance, and compensatory strategies (Cash, 2012). These strategies serve to maintain or strengthen positive appearance evaluations, and enable the individual to temporarily escape, reduce, or regulate appearance-related discomfort (Cash, 2012). Only a few qualitative studies have explored these concepts in relation to CFAs. In one such study, older adults with CL/P discussed the importance of managing the ‘upkeep’ of their appearance in order to minimize social reactions to their visible difference (Hamlet & Harcourt, 2014). In a second study, efforts to evade negative social reactions were also associated with decisions to undergo further invasive treatment (Bemmels et al., 2013). Some concealment and compensatory strategies were also described by adults with CL/P as ways of distracting other people from their visible difference (Stock et al., 2015), including a reluctance to leave the house without wearing make-up, and a tendency to exaggerate their personality in social situations. The exaggeration of personality as a coping strategy to reduce discomfort in social settings was also described in a recent case-study (Tevik & Feragen, 2015). Further, adults have described the difficult and disorienting task of re-adjusting to a new appearance after surgery (Cadogan & Bennun, 2011). Finally,
coping strategies and resilience have been described as protecting subjective appearance evaluations by adults with CFAs (Beaune et al., 2004; Cadogan & Bennun, 2011).

Quantitative studies of young people with CL/P have reported an association between dissatisfaction with appearance and behavioural problems (Wehby et al., 2012), and with general adjustment (Berger & Dalton, 2011; Thomas, Turner, Rumsey, & Dowell, 1997). However, potential confounders, such as an underlying developmental delay could explain this association, and require further investigation (Wehby et al., 2012). Several other studies also have explored associations between satisfaction with appearance and different domains of psychological adjustment (see Feragen & Stock, 2017; Stock & Feragen, 2016). However, again, most studies are based on cross-sectional samples that do not allow for an interpretation of directionality, calling for future research to utilize longitudinal designs.

Discussion

Synthesis of findings

This article has summarised the literature investigating subjective satisfaction with appearance among individuals affected by a CFA, in order to explore factors that could explain the development and maintenance of, and variation in, subjective appearance evaluations. For the first time, the cognitive-behavioural model of body image (Cash, 2012) has been applied to findings from the CFA field, in order to address potential similarities between the two research areas, to identify potential gaps in the craniofacial literature, and to provide orientation for future research across both fields.

Cash’s model (2012) consists of predisposing and proximal factors thought to affect and maintain individuals’ subjective evaluations of body image (Figure 1). In applying this model to the craniofacial literature, a number of comparable features are apparent. Specifically, much work has been done in relation to interpersonal experiences and its association with satisfaction with appearance,
particularly in regard to social reactions to looking ‘different’, appearance-related teasing, and positive social experiences as a potential buffer for appearance dissatisfaction. Similarly, several associations between satisfaction with appearance and fear of negative evaluation, body image emotions, and emotional distress have been observed within the CFA literature. Many studies have investigated appearance evaluations in relation to CFAs, with the assumption that unusual physical characteristics will lead to more appearance dissatisfaction. While there is some support for this, internal psychological factors have been shown to be much stronger predictors of appearance satisfaction than physical characteristics alone. Thus, a call has been made to investigate the psychological factors which may lead to or perpetuate appearance dissatisfaction, rather than simply noting the presence of appearance satisfaction or dissatisfaction within CFA samples (Stock et al., in press). Finally, two activating events have been investigated in the context of CFAs, and seem to fit well with Cash’s 2012 model (Figure 1). Negative social experiences may draw attention to the visible difference, and lead to the affected features becoming a focus for appearance dissatisfaction. While appearance may change naturally over the lifespan, those with CFAs are also likely to undergo several appearance-altering surgeries during their lifetime, adding an additional element to the model.

The application of Cash’s model (Figure 1) to the available craniofacial literature has also highlighted a number of gaps in current theoretical knowledge in the field. Those affected by CFAs arguably differ not only from the ‘ideal’ appearance, but also from the ‘norm’. There is much work surrounding Western societal attitudes to those with an unusual appearance, but little on how this is related to the affected individual’s subjective satisfaction with their appearance, particularly across cultures, indicating a gap in current understanding. There has also been very little work into personality factors among individuals born with a CFA, particularly in recent years, and specifically how this may relate to appearance satisfaction. In the last few decades, there seems to be a shift from searching for pathological personality dimensions that could be a consequence of living with an unusual appearance, toward a more ‘normalising’ approach and a search for individual characteristics that could protect the
individual against negative social experiences. Finally, there is a limited amount of literature available in relation to appearance investment, the importance of appearance, and self-regulatory behaviours among those with CFAs. Where internal cognitive factors have been investigated, this work has focused on possible protective factors, and potential ways of facilitating resilience among those with a visible difference; an approach which may also be beneficial for the general population. Last, an interesting difference in appearance-related terminology used in the CFA research, and in the more general body image literature was evident. While the craniofacial literature has predominantly focused on terms such as ‘subjective appearance evaluations’ or ‘appearance (dis)satisfaction’, less focus has been given on other components of body image definitions.

**Methodological considerations**

This article has synthesized the available literature pertaining to appearance satisfaction in the context of CFAs. The potential impact of demographic factors has not been discussed, since these have been extensively described in previous reviews (see Feragen & Stock, 2017; Hunt et al., 2005; Stock & Feragen, 2016). Only papers investigating factors that were found to be related to the individual’s subjective satisfaction with appearance were included. Further, papers discussing their findings in terms of their associations with appearance were only included if appearance was explicitly described as one of the study outcomes. One challenge with this was that many qualitative papers, by nature, did not necessarily present findings as being specifically related to appearance satisfaction, making some of the findings more difficult to tease apart. This is indicative of the implicit assumption that appearance is an included variable simply because the sample of interest is affected by a visible difference; a challenge that was evident both in the qualitative and in the quantitative literature. Again, there is a need to investigate the psychological factors which may facilitate and prevent appearance dissatisfaction, beyond the presence or absence of a visible difference.
This paper has summarised and discussed the available literature regarding CFAs, drawing on literature from the wider field of visible difference for contextual purposes where appropriate. However, research relating specifically to other conditions affecting appearance (such as a birthmark, burns injury, skin condition or an altered appearance due to cancer treatment) was not included. Thus, there is a need for future studies to investigate the potential validity of this paper’s conclusions in relation to other visible conditions.

As outlined in Cash’s cognitive-behavioural model of body image development (2012; Figure 1), the differentiation between predisposing historical factors and current proximal events can be difficult to draw. In some cases, it can be difficult to differentiate between the various events and factors, and therefore some aspects may be appropriately situated under more than one subheading. This was specifically challenging in relation to the qualitative literature, where the direction of variables may be less explicitly described. In addition, the use of concepts may vary and sometimes be unclear within the qualitative literature, while the use of outcome measures facilitates the reader’s understanding of included variables in quantitative studies. A positive consequence of these challenges was that included qualitative studies could shed light on several aspects of the cognitive-behavioural model, while quantitative studies usually only investigated one or two aspects of appearance. The synthesis of studies exploring factors affecting subjective satisfaction with appearance is also complicated by the high variation in methods and measures used in CFA research (Klassen et al., 2012). There is a need for future studies to agree upon conceptual frameworks and measures, and to draw upon broader literature (Stock et al., in press), in order to accelerate understanding of the complex nature of psychological adjustment to living with a facial difference.

Conclusions

Although additional research is necessary, Cash’s cognitive-behavioural model of body image development (2012) appears to fit relatively well with the existing craniofacial literature. Since there is
little evidence of psychological theories or models being applied to CFA research findings (Baker et al., 2009; Stock et al., in press), the present paper hopes to make a useful contribution in addressing this methodological concern. This paper has also highlighted a number of weaker areas within the CFA field, which may benefit from future investigation, and has highlighted the need for more longitudinal research in order to establish the directionality of associations. Finally, the literature outlined in this article suggests considerable overlap between factors of interest in the field of CFAs, and in the area of body image more broadly, in spite of an evident variation in the use of appearance-related terminology. A closer collaboration between general body image research, and research into the impact of a congenital visible difference could therefore offer a more comprehensive picture of how body image concerns are developed and maintained, as well as how to implement prevention and intervention strategies across both fields.
References


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