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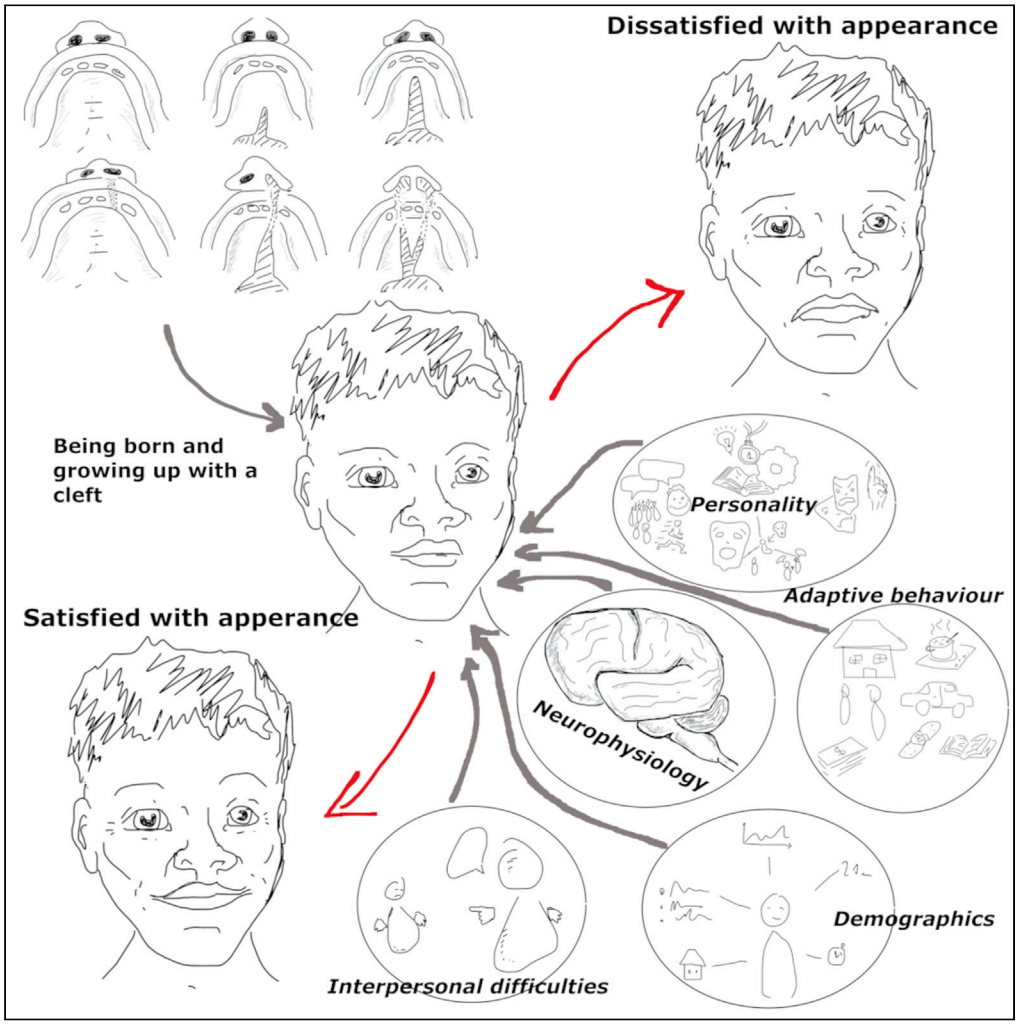

Jakob Sajovic, Gorazd Drevenšek, Alja Plut, ..., Anina Setnikar Lesjak, Polona Selič Zupančič, Martina Drevenšek

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Highlights

Interpersonal difficulties negatively impact satisfaction with appearance

Conscientiousness positively influences satisfaction with appearance across models

Frontal alpha asymmetry explains satisfaction, but the interaction is complex

Older age is associated with lower satisfaction with appearance in young people

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Beyond the face: An interdisciplinary evaluation of satisfaction with appearance in young people with orofacial clefts

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SUMMARY

Orofacial clefts are the most common congenital anomaly of the face, and they significantly affect appearance. The combined effects of demographics, psychology, neurophysiology, and cleft characteristics to explain satisfaction with appearance in young people with a cleft have not yet been comprehensively studied in an interdisciplinary manner. We found that interpersonal difficulties, age, and conscientiousness were significant explanatory factors for satisfaction with appearance ($t_{interpersonal\ difficulties} = -3.022$, $p = 0.006$; $t_{age} = -3.563$, $p = 0.016$; $t_{conscientiousness} = 4.161$, $p = 0.003$); the model explained 50% of variance in satisfaction with appearance ($R^2_{Adjusted} = 0.504$, $F_{vs.\ constant} = 4.05$, $p = 0.00117$). Furthermore, frontal alpha asymmetry was complexly intertwined with other variables, affecting the overall accuracy of the model, but explaining only 10.5% of variance in satisfaction with appearance when used as a factor alone. The results show that an interdisciplinary approach can substantially expand our understanding of the factors influencing self-perception in young people with orofacial clefts.

INTRODUCTION

Orofacial clefts are the most prevalent congenital anomaly of the craniofacial complex, affecting roughly 1 in 1,000 live births worldwide.[1,2] They develop between the fourth and tenth week of gestation,[2] and can affect the primary palate (lip and/or alveolus), the secondary palate (the roof of the mouth) or both; can be unilateral (affecting only one side of the face) or bilateral (affecting both sides of the face); and can be complete or incomplete (from a small notch on the upper lip to a complete defect involving the nostrils, or from a mild submucous cleft to a complete cleft of the soft and hard palate).[1–3]

A classification of the type of cleft into primary palate cleft, secondary palate cleft, and primary and secondary palate cleft is often used, as clefts of the primary and secondary palate have different developmental pathways, and because they give rise to different difficulties in the affected child.[1] Etiologically, the most important distinction is between syndromic and non-syndromic clefts, distinguishing between those which are a result of a broader genetic disorder, where one of the consequences of the disorder is a cleft, but other symptoms are also present; and those where the cleft is the main symptom, without other connected issues and without necessary genetic involvement.[1,2]

As an orofacial cleft affects the face, the first part of the body that others notice, it not only significantly affects the appearance of people born with it, but can also affect their lives in other ways.[4,5] These effects are particularly pronounced in young people, starting at the beginning of puberty and continuing into early adulthood.[6] Studies consistently report lower health-related quality of life, poorer emotional well-being, and lower self-esteem in young people with cleft lip and/or palate compared to healthy peers.[7,8] Young people with a cleft experience more frequent negative emotions like anger, sadness, and fear, and report feeling alienated from their peers[8]; they also exhibit higher levels of social anxiety, more behavioral problems, and more frequent symptoms of depression.[8,9]

Crucially, lower satisfaction with appearance and speech are strongly associated with a higher frequency of emotional problems in young people with a cleft lip and/or palate.[10,11] These problems are often clinically significant, exceeding those observed in healthy peers.[12] However, the impact of a cleft on psychosocial adjustment varies with gender, age, and specific cleft diagnosis.[5,8,13] Poorer psychosocial adjustment and dissatisfaction with appearance or speech significantly hinder social relationships for young people with a cleft.[8] This is further compounded by social stigma associated with their altered appearance and potential communication difficulties, which may negatively impact academic performance.[13]

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Interestingly, research also suggests potential positive consequences of having a cleft.[11,14–16] Some studies have reported increased development of psychosocial skills, personal resilience, appreciation of diversity, and better coping abilities in young people with a cleft compared to unaffected peers.[11,14,15] It has also been shown that satisfaction with appearance may be better in young people with a cleft than in the general population from childhood to early adulthood, especially when aspects unaffected by having a cleft are concerned.[16] These conflicting findings may be explained by hidden factors which differ in those born with a cleft and those born without, such as social skills, resilience or optimism.[17]

Satisfaction with appearance in young people with a cleft has been investigated in conjunction with a variety of variables and characteristics of young people with a cleft, including demographics,[18,19] psychological and psychosocial variables,[8,20–22] and surgical and orthodontic treatment approaches, in combination with demographics, cleft type, gender, and parental characteristics,[23–25] with at most two such viewpoints utilized at once.[8,18,19,21,22,24] While previous research has identified the importance of psychological characteristics, personality, demographics, and cleft-related features on appearance satisfaction in young people with a cleft,[8,18,19,21,22,24] no prior studies have attempted an interdisciplinary evaluation of these factors to understand their combined influence.

As has been shown, various psychological, demographic and clinical characteristics of people with a cleft are complexly intertwined with satisfaction with appearance.[18,19] In this study, a thorough multidimensional investigation of the interaction of these variables was conducted, aiming to aid the understanding of satisfaction with appearance embedded in the complex interplay of factors. The use of self-report measures in assessing psychological characteristics, clinical outcomes from the viewpoint of the patient, and satisfaction with appearance was augmented (but not replaced), by appropriate physiological measurements. Electroencephalography (EEG) as an adjacent measure to the psychological characteristics of young people with a cleft was included in the present study, as it has previously been shown that measures derived from it can be good correlates of emotion[26,27] and personality characteristics.[28] Frontal alpha asymmetry (8-13 Hz) at rest, a reasonably well-studied marker of emotional states,[26,28] was utilized, given that higher right frontal alpha power is known to be associated with negative emotions such as trait anxiety, depression, and low self-esteem.[26,28,29] Reduced alpha power has also been linked to stress.[27] While informative on both temperament and mood, frontal alpha asymmetry is not specific to either.[26–29] It has also recently been associated with the big five personality traits, if the EEG data is carefully pre-processed.[28]

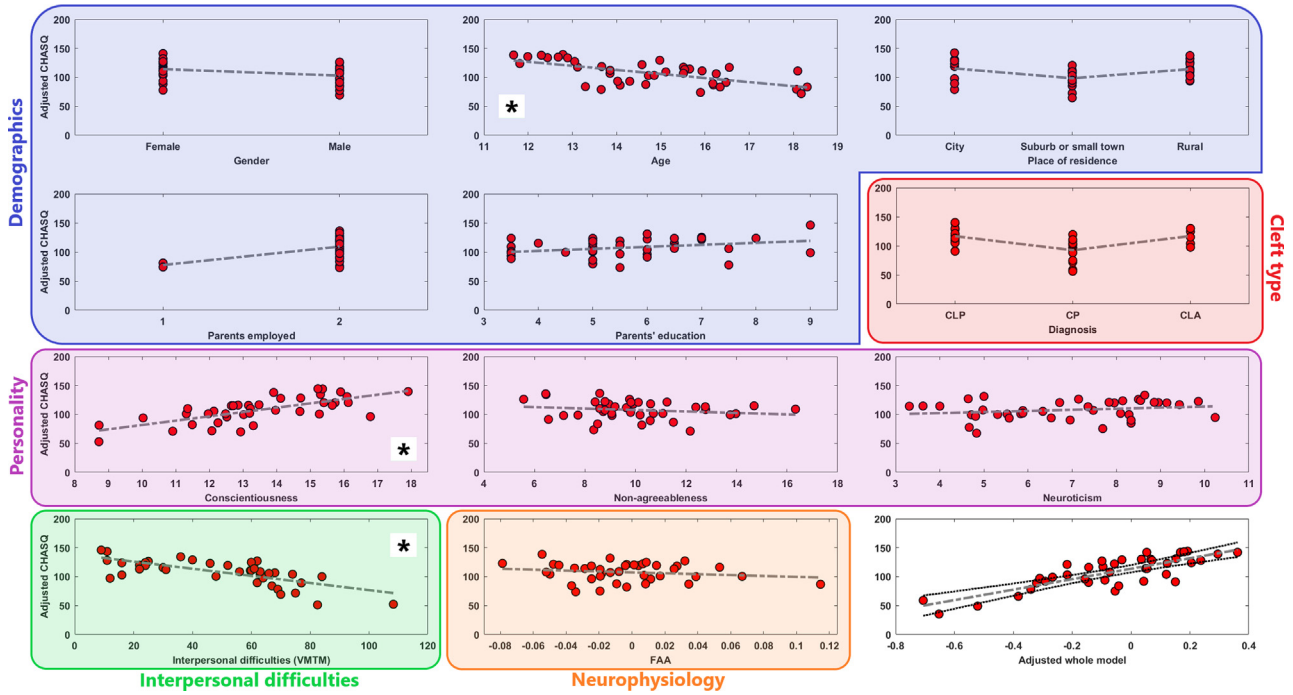


Figure 1. An adjusted partial regression plot for the model testing the effects of frontal alpha asymmetry (FAA), neuroticism and interpersonal difficulties (VMTM all) on satisfaction with appearance in young people with a cleft (CHASQ), while controlling for the effects of other psychological, socio-economic, and demographic variables

The plots show the adjusted effects of each variable included in the model, meaning that all influence of the other variables included in the model has been removed, showing the isolated effect of the variable depicted. Red circles with black borders represent individual data points ($F_{vs.\ constant} = 4.05$, df = 26, $p = 0.00117$, $N = 40$ participants), also adjusted to eliminate the effects of other variables. The gray dash-and-dot lines represent the adjusted regression fit for each variable. The bottom right panel shows the adjusted plot for the entire model. There, the black dashed lines represent the 95% confidence interval for the whole-model regression fit (dash-and-dot gray line). The variables are grouped into categorical (marked with a blue surface), cleft type (marked with a red surface), personality (marked with a lavender surface), interpersonal difficulties (marked with a light green surface) and neurophysiology (orange surface). The non-colored surface covered graph is the adjusted whole model. All plots have the adjusted CHASQ values as their y axis. An * marks a significant effect.

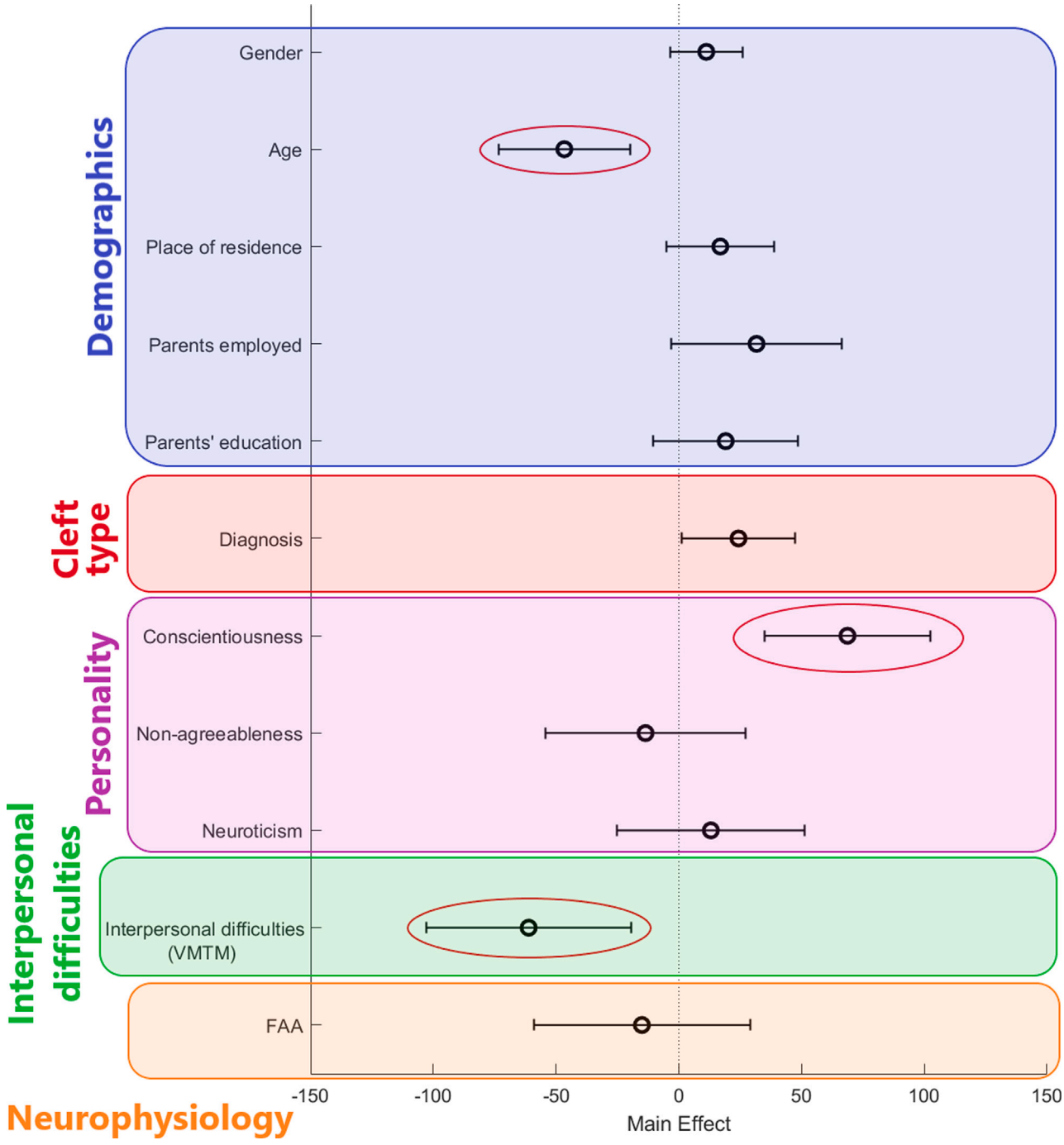


Figure 2. This figure depicts the main effects of the regression model testing the effects of frontal alpha asymmetry, neuroticism and interpersonal difficulties on their satisfaction with appearance, while controlling for the effects of other psychological, socio-economic and demographic variables ($F_{vs.\ constant}$ = 4.05, df = 26, p = 0.00117, N = 40 participants)

The main effect (x axis) shows the total isolated effect of each variable (black circles), when all other variables in the model have been controlled for. This means that the effects depicted show how much the entire span of the variable has affected satisfaction with appearance. The black capped lines represent 95% confidence intervals for the effects. Red ellipses represent significant effects, at the α = 0.05 level, after the Bonferroni correction was applied. Please note that for this reason, diagnosis is not marked as significant, although its 95% confidence interval does not intersect with the dotted line. The variables are grouped into demographic (marked with a blue surface), cleft type (marked with a red surface), personality (marked with a lavender surface), interpersonal difficulties (marked with a light green surface) and neurophysiology (orange surface). FAA = frontal alpha asymmetry.

Table 1. Results of the fitting of the linear regression model for the data of subjects with electroencephalography data available

	Coefficient estimate	SE	t-statistic	*p*-value	Bonferroni corrected *p*-value
Intercept of the model	82.043	62.777	1.307	0.203	Not applicable
Neuroticism	1.883	2.674	0.704	0.488	Not applicable
Interpersonal difficulties	−0.616	0.204	−3.022	***0.006***	Not applicable
Frontal alpha asymmetry	−77.783	110.658	−0.703	0.488	Not applicable
Gender (female vs. male)	−11.201	7.121	−1.573	0.128	1.000
Age	−7.019	1.970	−3.563	**0.001**	**0.016**
Place of residence – suburb vs. city	−16.891	10.684	−1.581	0.126	1.000
Place of residence – rural vs. city	−1.108	9.466	−0.117	0.908	1.000
Conscientiousness	7.470	1.795	4.161	**0.000**	**0.003**
Non-agreeableness	−1.262	1.834	−0.688	0.497	1.000
Diagnosis – CLP vs. CP	−24.088	9.755	−2.469	**0.020**	0.225
Diagnosis – CLP vs. CLA	0.220	11.193	0.020	0.984	1.000
Parents' education	3.470	2.617	1.326	0.196	1.000
Parents' employment	31.651	16.977	1.864	0.074	0.810

CLP, cleft lip and palate; CP, cleft palate; CLA, cleft lip and alveolus. In italic script are the factors tested *a-priori*. The first row is the intercept.

This study therefore aimed to provide the first foray into interdisciplinary investigation of the combined effects of psychological, social, neurophysiological, demographic, and clinical factors on the satisfaction with appearance of young people with a cleft lip and/or palate.

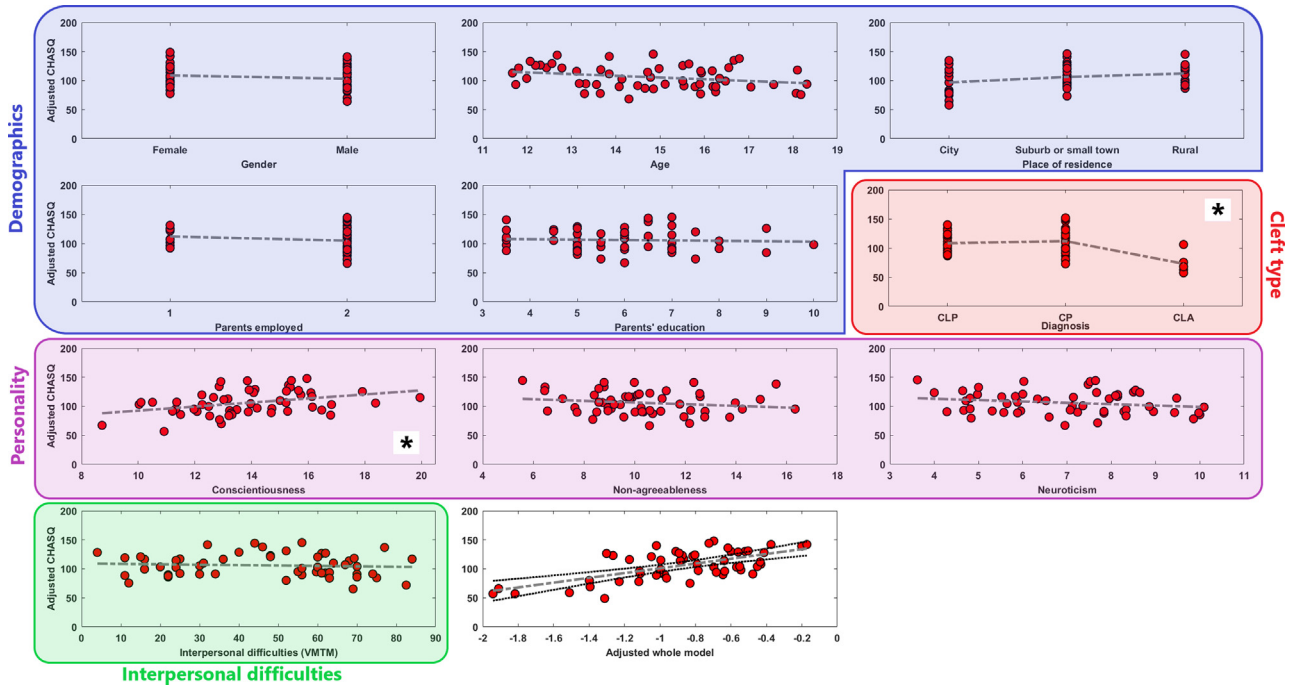


Figure 3. An adjusted partial regression plot for the model testing the effects of neuroticism and interpersonal difficulties (VMTM all) on satisfaction with appearance of young people with a cleft (CHASQ), while controlling for the effects of other psychological, socio-economic, and demographic variables on an extended sample of young people with a cleft ($F_{vs.\ constant}$ = 2.52, df = 42, p = 0.0136, N = 55 participants)

The plots show the adjusted effects of each variable included in the model, meaning that all influence of the other variables included in the model has been removed, showing the isolated effect of the variable depicted. Red circles with black borders represent individual data points, also adjusted to eliminate the effects of other variables. The gray dash-and-dot lines represent the adjusted regression fit for each variable. The bottom right panel shows the adjusted plot for the entire model. There, the black dashed lines represent the 95% confidence interval for the whole-model regression fit (dash-and-dot gray line). The variables are grouped into categorical (marked with a blue surface), cleft type (marked with a red surface), personality (marked with a lavender surface) and interpersonal difficulties (marked with a light green surface). The non-colored surface covered graph is the adjusted whole model. All plots have the adjusted CHASQ values as their y axis. An * marks a significant effect.

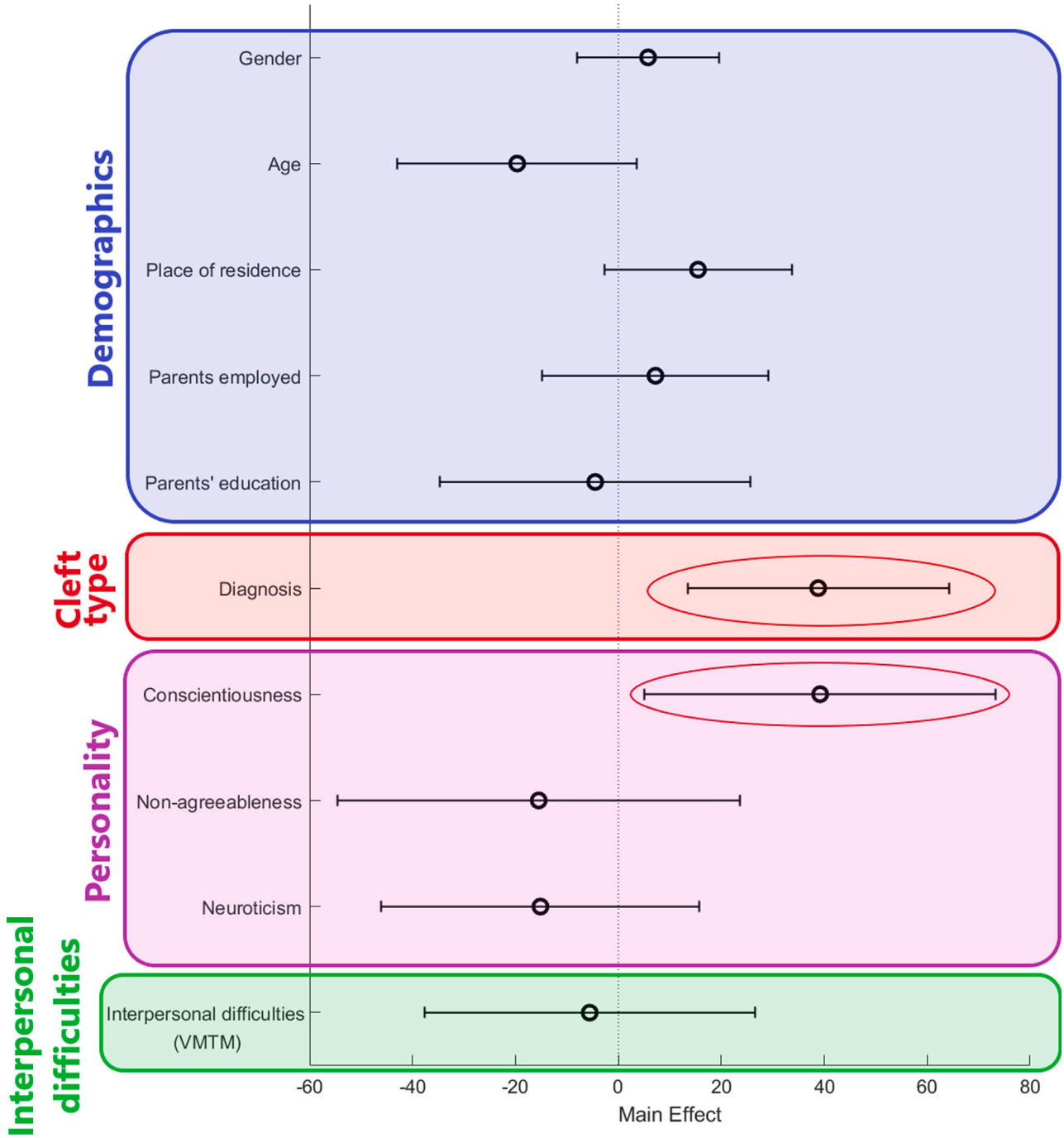


Figure 4. This figure depicts the main effects of the regression model testing the effects of frontal alpha asymmetry, neuroticism and interpersonal difficulties on their satisfaction with appearance, while controlling for the effects of other psychological, socio-economic and demographic variables ($F_{vs.\ constant} = 2.52$, df = 42, $p = 0.0136$, $N = 55$ participants)

The main effect (x axis) shows the total isolated effect of each variable (black circles), when all other variables in the model have been controlled for. This means that the effects depicted show how much the entire span of the variable has affected satisfaction with appearance. The black capped lines represent 95% confidence intervals for the effects. Red ellipses represent significant effects, at the $\alpha = 0.05$ level, after the Bonferroni correction was applied. The variables are grouped into demographic (marked with a blue surface), cleft type (marked with a red surface), personality (marked with a lavender surface) and interpersonal difficulties (marked with a light green surface). FAA = frontal alpha asymmetry.

Table 2. Results of the fitting of the linear regression model for the data of subjects without electroencephalography data available

	Coefficient	SE	t-value	*p*-value
Intercept of the model	149.293	49.579	3.011	**0.004**
Neuroticism	−2.340	2.368	−0.988	0.329
Interpersonal difficulties	−0.070	0.199	−0.351	0.728
Place of residence – suburb vs. city	9.475	8.349	1.135	0.263
Place of residence – rural vs. city	15.466	9.028	1.713	0.094
Conscientiousness	3.493	1.507	2.317	**0.025**
Non-agreeableness	−1.439	1.804	−0.798	0.430
Diagnosis – CLP vs. CP	3.627	7.622	0.476	0.637
Diagnosis – CLP vs. CLA	−35.196	12.041	−2.923	**0.006**
Parents' education	−0.693	2.296	−0.302	0.764
Parents' employment	−7.205	10.884	−0.662	0.512
Gender	−5.761	6.832	−0.843	0.404
Age	−2.961	1.736	−1.706	0.095

CLP, cleft lip and palate; CP, cleft palate; CLA, cleft lip and alveolus. The first row shows the intercept.

We hypothesized that young people who experience greater difficulties interacting with others (interpersonal difficulties), tend to worry more (higher neuroticism), and have trouble adjusting socially (poorer social adaptive behavior) would have lower satisfaction with their appearance. Additionally, we expected that young people with a greater right frontal alpha asymmetry would also report lower satisfaction with their appearance.

RESULTS

Age, conscientiousness, and interpersonal difficulties are significant explanatory factors of satisfaction with appearance (Figures 1 and 2; Table 1). The variables included in the model were gender, age, place of residence, conscientiousness, non-agreeableness, neuroticism, interpersonal difficulties, diagnosis, parents' education, parents' employment, and frontal alpha asymmetry; adjusted partial regression plots for each are shown in Figure 1.

The main effects of gender, age, place of residence, conscientiousness, non-agreeableness, neuroticism, interpersonal difficulties, cleft diagnosis, parental education, parental employment, and frontal alpha asymmetry, are shown in Figure 2.

The coefficients and their standard errors, t-statistics and *p*-values for the model in Figures 1 and 2 are presented in Table 1. Significant results are presented in bold font.

The number of observations included in the model = 40; error degrees of freedom = 26; the root mean squared error = 19.1; $R^2 = 0.67$; $R^2_{Adjusted} = 0.504$; $F_{vs.\ constant} = 4.05$; $p = 0.00117$.

From here on, the results of exploratory analyses are presented. Conscientiousness and diagnosis were the only significant explanatory factors of satisfaction with appearance when 17 new cases were added to the model and alpha asymmetry was removed as an explanatory factor (Figures 3 and 4; Table 2). The robustness of the observed conscientiousness effect was tested. Figure 3 shows the adjusted partial regression plots for gender, age, place of residence, conscientiousness, non-agreeableness, neuroticism, interpersonal difficulties, cleft diagnosis, education of the parents and employment of the parents in the extended sample model.

The main effects of gender, age, place of residence, conscientiousness, non-agreeableness, neuroticism, interpersonal difficulties, cleft diagnosis, parental education and parental employment, using the extended sample, are presented in Figure 4.

The coefficients, their standard errors, t-statistics, and *p*-values for the model presented in Figures 3 and 4 are listed in Table 2.

The number of observations in this model = 55; error degrees of freedom = 42; root mean squared error = 21.7; $R^2 = 0.418$; $R^2_{Adjusted} = 0.252$; $F_{vs.\ constant} = 2.52$; *p*-value = 0.0136.

Conscientiousness and age were significant explanatory factors of satisfaction with appearance if alpha asymmetry was removed from the primary model (Figures 5 and 6). The effect of removing the alpha asymmetry from the model was tested. Figure 5 shows the adjusted partial regression plots for gender, age, place of residence, conscientiousness, non-agreeableness, neuroticism, interpersonal difficulties, cleft diagnosis, parental education and parental employment; the same variables as in the primary sample model, without alpha asymmetry.

The main effects of gender, age, place of residence, conscientiousness, non-agreeableness, neuroticism, interpersonal difficulties, cleft diagnosis, parental education and parental employment, using the same sample as the primary model without alpha asymmetry, are shown in Figure 6.

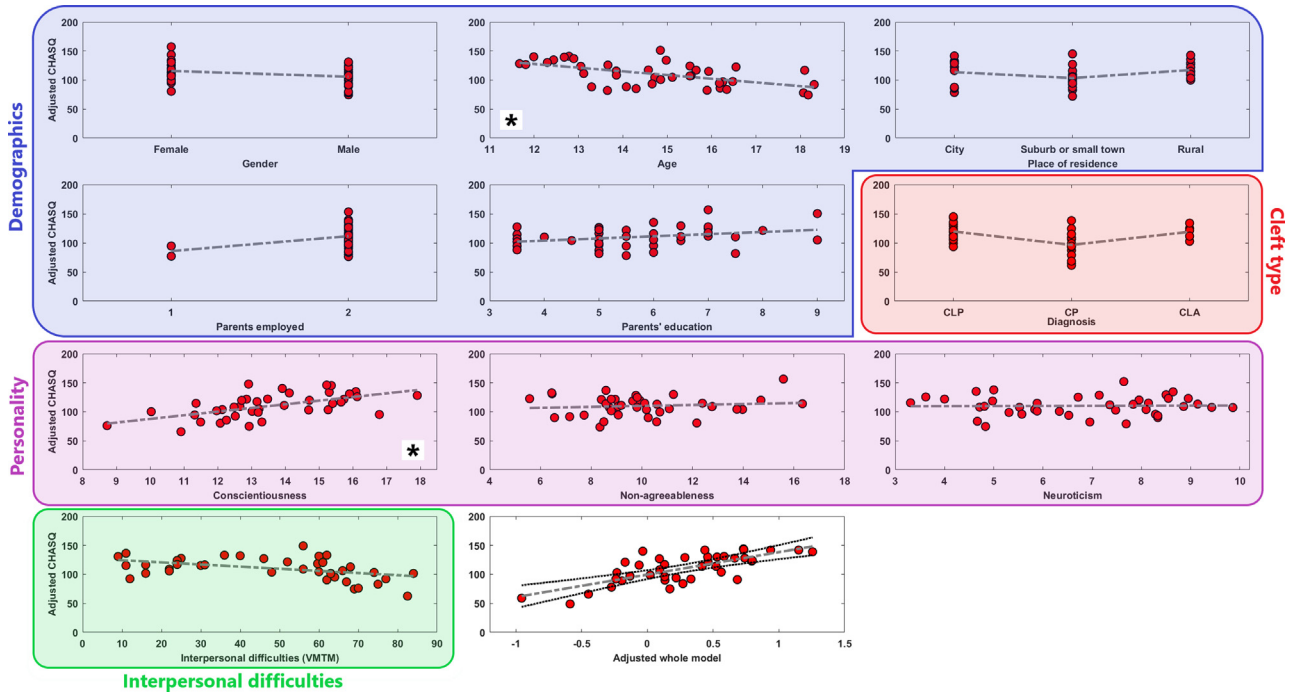


Figure 5. An adjusted partial regression plot for the model testing the effects of neuroticism and interpersonal difficulties (VMTM all) on satisfaction with appearance of young people with a cleft (CHASQ), while controlling for the effects of other psychological, socio-economic and demographic variables, and using the sample for which electroencephalography data is available but excluding this data from the model ($F_{vs.\ constant}$ = 2.77, df = 28, p = 0.0128, N = 41 participants)

The plots show the adjusted effects of each variable included in the model, meaning that all influence of the other variables included in the model has been removed, showing the isolated effect of the variable depicted. Red circles with black borders represent individual data points, also adjusted to eliminate the effects of other variables. The gray dash-and-dot lines represent the adjusted regression fit for each variable. The bottom right panel shows the adjusted plot for the entire model. There, the black dashed lines represent the 95% confidence interval for the whole-model regression fit (dash-and-dot gray line). The variables are grouped into categorical (marked with a blue surface), cleft type (marked with a red surface), personality (marked with a lavender surface) and interpersonal difficulties (marked with a light green surface). The non-colored surface covered graph is the adjusted whole model. All plots have the adjusted CHASQ values as their y axis. An * marks a significant effect.

The number of observations = 41; error degrees of freedom = 28; root mean squared error = 21.8; R^2 = 0.543; $R^2_{Adjusted}$ = 0.347; $F_{vs.\ constant}$ = 2.77; p = 0.0128.

Alpha asymmetry on its own was a significant explanatory factor of satisfaction with appearance (Figure 7). The independent effect of alpha asymmetry on satisfaction with appearance was tested.

In more detail, the number of observations for this model = 47; error degrees of freedom = 45; root mean squared error = 23.3; R^2 = 0.124; $R^2_{Adjusted}$ = 0.105; $F_{vs.\ constant}$ = 6.39; p = 0.015.

The results in Figure 8 show significant differences in the extent of interpersonal difficulties (F = 3.506; df = 2; p = 0.0358) between the young people with the three cleft subtypes, but no significant differences in satisfaction with appearance or conscientiousness.

DISCUSSION

This study presents a novel approach to understanding the interplay between the psychological characteristics, social factors, clinical evaluations, and neurophysiological characteristics which shape satisfaction with appearance in young people with an orofacial cleft, by employing an interdisciplinary framework. Our findings confirmed the significant effect of interpersonal difficulties (Figures 1 and 2; Table 1) but, surprisingly, reveal conscientiousness as another key explanatory factor (Figures 1, 2, 3, 4, 5, and 6; Tables 1 and 2). Additionally, frontal alpha asymmetry appeared to interact with other variables, suggesting a complex interplay which requires further exploration (Figures 3, 4, 5, 6, and 7).

The detrimental effect of interpersonal issues on satisfaction with appearance observed in our study (Figures 1 and 2; Table 1) aligns with previous research[30,31] and our initial hypothesis, and is not a result of exploratory analysis, thus making this effect the best grounded of all the findings in the present study.

This finding can be explained by interpersonal difficulties, negative self-perception and poorer satisfaction with appearance being linked and developing together.[32] The pathways between them that influence satisfaction with appearance can work in two ways, more interpersonal

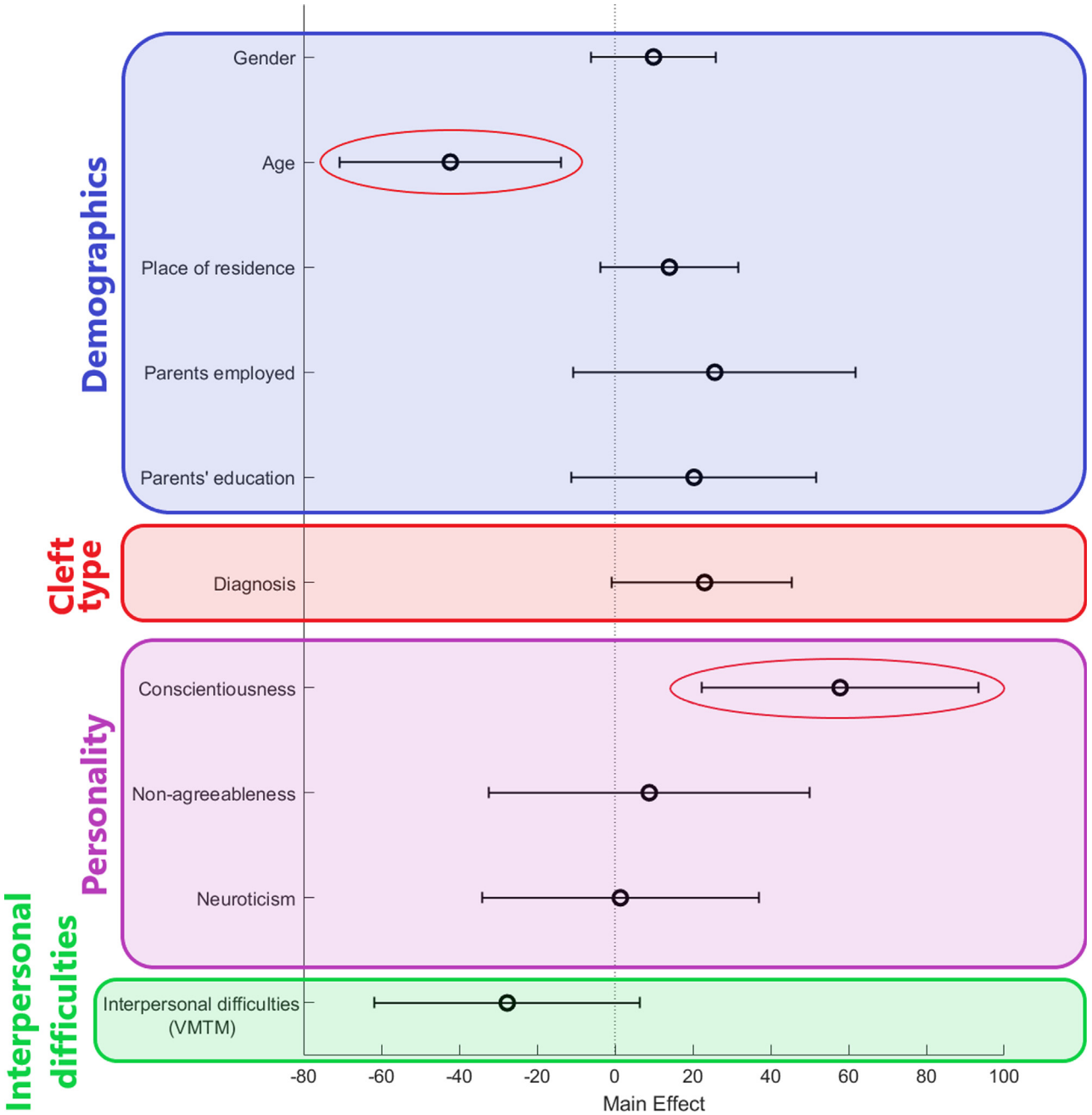


Figure 6. This figure depicts the main effects of the regression model testing the effects of neuroticism and interpersonal difficulties on their satisfaction with appearance, while controlling for the effects of other psychological, socio-economic and demographic variables

The main effect (x axis) shows the total isolated effect of each variable (black circles), when all other variables in the model have been controlled for ($F_{vs.\ constant}$ = 2.77, df = 28, p = 0.0128, N = 41 participants). This means that the effects depicted show how much the entire span of the variable has affected satisfaction with appearance. The black capped lines represent 95% confidence intervals for the effects. Red ellipses represent significant effects, at the α = 0.05 level, after the Bonferroni correction was applied. The variables are grouped into demographic (marked with a blue surface), cleft type (marked with a red surface), personality (marked with a lavender surface) and interpersonal difficulties (marked with a light green surface). FAA = frontal alpha asymmetry.

difficulties resulting in negative self-perception and poorer satisfaction with appearance, or poorer satisfaction with appearance influencing self-perception and the social interactions of young people, resulting in more interpersonal difficulties.[30,31] The mechanisms involved should be studied in more detail in purposely designed studies.

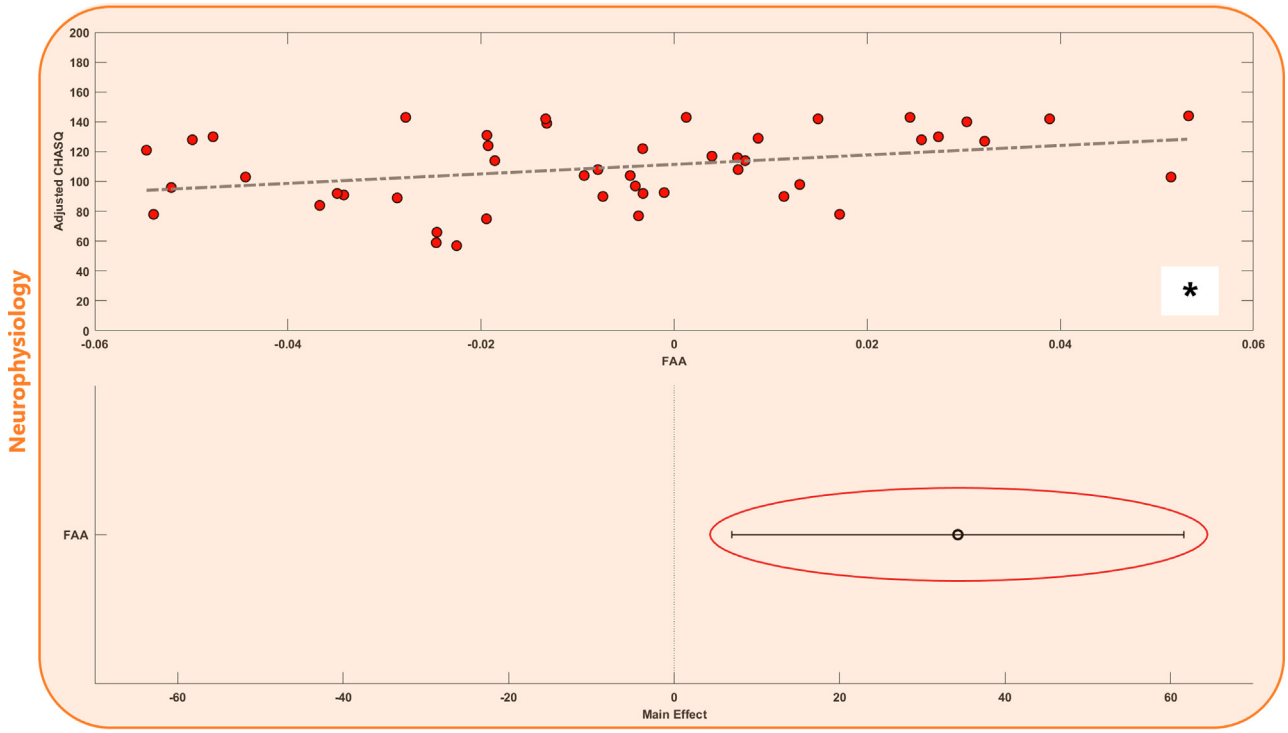


Figure 7. The results of univariate regression of frontal alpha asymmetry (FAA) to satisfaction with appearance in young people with a cleft (CHASQ)
The top panel shows the adjusted partial regression plot without the effects of the intercept; the blue line represents the regression fit and the red dots the individual data points ($F_{vs.\ constant}$ = 6.39, df = 45, p = 0.015, N = 47 participants). The bottom panel shows the main effect plot, with the red ellipse indicating significance. The orange color shows that this variable represents neurophysiology of young people with clefts.

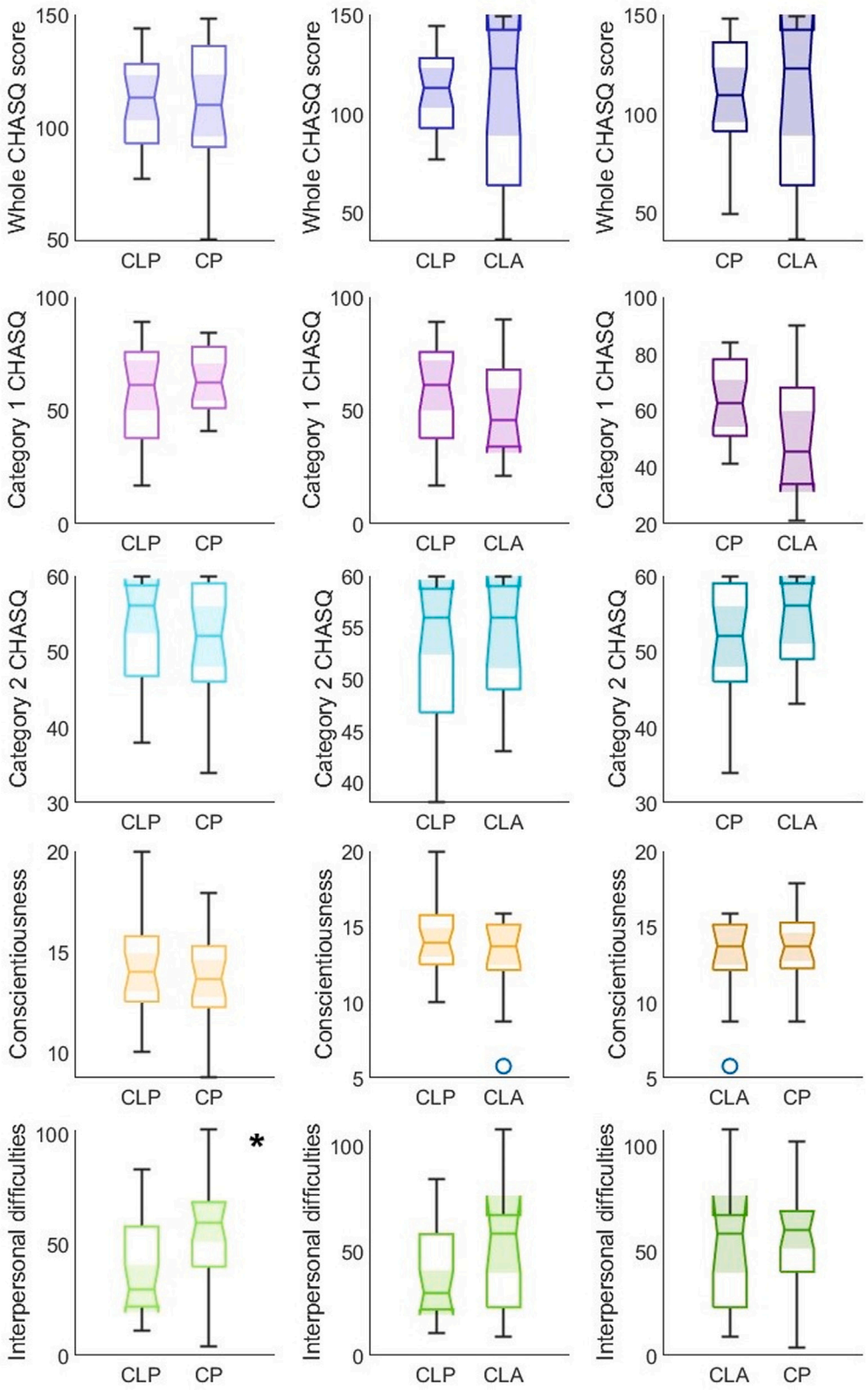
Frontal alpha asymmetry was found to capture similar facets of satisfaction with appearance as other variables analyzed in this study (Figures 1, 2, 3, 4, 5, 6, and 7; Tables 1 and 2). At first, we expected greater right frontal alpha asymmetry to be associated with decreased satisfaction with appearance, but this assumption was not supported when considering the influence of personality traits, social factors, and other specific characteristics of young people with a cleft (Figures 1 and 2; Table 1). When conducting exploratory analyses to better determine how conscientiousness is tied to satisfaction with appearance, we noticed that that the removal of alpha asymmetry changes the characteristics of our model significantly (Figures 3, 4, 5, and 6; Table 2). Exploring further by evaluating alpha asymmetry independently of the other variables, we discovered that it does emerge as a significant explanatory factor of appearance satisfaction, albeit not a particularly strong one, accounting for only 10.5% of the variance in satisfaction levels (Figure 7).

Although any conclusions on this topic are doubtlessly tentative and should be confirmed or rejected in future research, we provide a possible explanation for the observed pattern of results regarding the frontal alpha asymmetry. A possible explanation is that right frontal alpha asymmetry is associated with self-esteem, as this has been observed in a previous study,[29] which in turn is associated with satisfaction with appearance.[33,34] This association may explain why alpha asymmetry appeared to positively correlate with appearance satisfaction when it is the sole factor under consideration (Figure 7). Yet, when alpha asymmetry was analyzed alongside other variables (Figures 1 and 2; Table 1), its explanatory power diminished and its relationship with appearance satisfaction inverted to negative, possibly reflecting more of the supposed approach-avoidance motivation aspects.[35] Furthermore, the removal of alpha asymmetry from our primary model led to a significant 15.7% decrease in the explained variance. This suggests that alpha asymmetry's role is multifaceted, entangled with a web of psychological, social, and demographic factors, and thus, it should not be simplistically viewed as a mere indicator of approach or avoidance motivation.[35] Rather, it should be considered within the broader social, psychological, and situational contexts of the individuals studied to fully understand its impact.

The most striking and unexpected finding was that conscientiousness had a significant positive effect on satisfaction with appearance (Figures 1, 2, 3, 4, 5, and 6; Tables 1 and 2), a finding reflected in both our primary model (Figures 1 and 2; Table 1) and exploratory analyses (Figures 3, 4, 5, and 6; Table 2). Additionally, it was also the explanatory factor the least affected by the removal of either alpha asymmetry from the model (Figures 5 and 6) or by the addition of more data (Figures 3 and 4; Table 2).

No prior association was established between conscientiousness and satisfaction with appearance in individuals born with a cleft, even though conscientiousness has been linked to improved health outcomes[36–38] and enhanced self-esteem[39,40] in broader populations. To explain this phenomenon, we can consider the following three explanations:

First, the well-documented relationship between conscientiousness and positive health behaviors may suggest that individuals with higher levels of conscientiousness are more diligent in following treatment protocols, avoiding risky behaviors, managing stress effectively, and


Whole CHASQ score
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CP

Figure 8. The results of the post-hoc comparisons for the one-way ANOVA on the data on the CHASQ, conscientiousness and interpersonal difficulties of young people with a cleft (*N* = 70 participants)

An * symbol in the panel denotes a significant difference (Interpersonal difficulties, t = 2.103, df = 67, $p = 0.0392$). The plots depict the median, the lower and upper quartiles, any outliers (computed using the interquartile range), and the minimum and maximum values that were not outliers. The tapered, shaded notch serves as a visual indicator of the significance of the differences, where box charts whose notches do not overlap have different medians at the 5% significance level. CLP = cleft lip and palate, CP = cleft palate only, CLA = cleft lip and alveolus, CHASQ = Cleft Hearing, Appearance, and Speech Questionnaire.

maintaining healthy dietary and self-care routines.[36–38] For young people with a cleft who are more conscientious, this could demonstrate as better self-care and, consequently, greater satisfaction with their appearance. However, this does not imply a direct association between better clinical outcomes and appearance satisfaction due to better patient compliance, given that previous studies have shown a weak correlation between clinical or aesthetic outcomes and appearance satisfaction in people born with a cleft.[41,42]

Second, the investment in self-care and adherence to treatment regimens may lead to a psychological phenomenon known as the sunk-cost fallacy,[43] where the sheer effort and resources poured into treatment might bias individuals to view their appearance more favorably. This interpretation does not suggest that such satisfaction is not genuine, but rather that it stems from internal motivational and cognitive factors, which coincides with several recent proposals to utilize cognitive biases to improve patient adherence to treatment.[44,45]

Finally, the third explanation considers the higher incidence of adverse emotional outcomes[19,20,46] and lower conscientiousness[46] observed in young people with a cleft compared to the general population. In this scenario, conscientiousness could be tied to depression,[47] with lower levels of conscientiousness and lower satisfaction with appearance being reflective of underlying symptoms of depression.[48,49]

Lower satisfaction with appearance was found to be associated with older age (Figures 1 and 2; Table 1). This effect of age can be elucidated by the trajectory of self-esteem and overall well-being observed in individuals born with a cleft.[6,31] Previous research has highlighted the myriad challenges faced by individuals with clefts during childhood and adolescence, including negative social interactions,[31] the burden of undergoing numerous medical procedures, and the stigma attached to their condition.[6] However, by adulthood, many of these difficulties tend to diminish, leading to improvements in self-esteem and improved psychological state.[31]

Given that the participants in our study were all adolescents (See the STAR methods section, STAR Table A), it is reasonable to infer that they were likely still grappling with many of these challenges, with the resulting stress compounding and explaining the negative impact of age. Furthermore, based on the anticipated trajectory of improvement into adulthood, we might expect a reversal of this trend shortly after the age range of our subjects. However, further research endeavors are needed to clarify this.

The third set of exploratory findings shows that the CP group faced more interpersonal challenges than their CLP counterparts (Figure 8). Notably, no discernible differences were observed between the CLA group and either the CP or CLP groups. Our objective with this stream of analysis was to determine whether young people with different types of clefts differed in our main outcome measure, as some group differences emerged as potentially significant factors in other exploratory analyses (Figures 3, 4, 5, and 6; Table 2), and in the two main explanatory factors (interpersonal difficulties and conscientiousness) for satisfaction with appearance. This finding suggests a potential vulnerability among young people with CP, possibly stemming from challenges related to speech impairments.[30]

This study combines clinical evaluation, psychology, and neuroscience to explore appearance satisfaction in young people with cleft conditions. Interpersonal difficulties emerged as a significant explanatory factor, while conscientiousness, beyond being a personality trait, was also central and stable in explaining how young people differ in satisfaction with appearance – a link previously unestablished in this demographic. The relationship between conscientiousness and satisfaction with appearance warrants further investigation, exploring the interplay between depression, conscientiousness and neurophysiology, to illuminate potential pathways for integrating psychological support into treatment protocols for young people with a cleft. Currently, the best supported approach to integrating these findings into clinical practice would be the implementation of cognitive-behavioral psychotherapy to address either depression, low conscientiousness or detrimental effects of interpersonal issues as standard adjunct therapy to current modes of treatment.[50]

This research also innovates by incorporating neurophysiological data, revealing that while frontal alpha asymmetry does not directly explain appearance satisfaction well, it interacts with the psychosocial variables, underscoring the complex interplay between brain activity and young people's perception of their own appearance.

Overall, the outcomes of our investigation demonstrate that incorporating interdisciplinary data – spanning brain activity, psychological traits, and demographic information – can substantially refine our understanding of the dynamic interrelations between these factors. This holistic approach enables a more accurate depiction of the complex array of factors that mold how we see ourselves.

Limitations of the study

The exploratory results should be treated as post-hoc and interpreted with caution, as they are not as reliable as the *a-priori* planned analysis. Thus, they were treated less strictly than the main model, but should still provide a valuable springboard for future research and enable realistic effect size calculations required for an in-depth exploration of satisfaction with appearance in young people with a cleft. Moreover, we did not include any measures of orofacial morphology in our analyses, which may help to elucidate the relationship between satisfaction with appearance, clinical treatment outcomes, psychology, social factors, and demographics in young people with a cleft. Our sample size also precluded us from detecting medium or small effects; instead we were only able to detect effects upwards of $f^2 = 0.6$, which are very large. We also propose that the results obtained in this study be confirmed with longitudinal studies, to establish more of a causal link between our variables and satisfaction with appearance. As it stands, this research provides the first insights into a multidimensional analysis of satisfaction

with appearance in young people with clefts, but better generalizability of the findings could be achieved by longitudinal designs employing sufficient numbers of diverse participants to detect even small effects.

RESOURCE AVAILABILITY

Lead contact

Further information and requests for resources and reagents should be directed to and will be fulfilled by the lead contact, Jakob Sajovic (jakob.sajovic@kclj.si or jakob.sajovic@gmail.com).

Materials availability

This study did not generate new unique reagents.

Data and code availability

- All data collected in this study have been deposited at figshare and are publicly available as of the date of publication, under the DOI: https://doi.org/10.6084/m9.figshare.25562667.v1. DOIs are also listed in the key resources table.
- All original code has been deposited at figshare and is publicly available as of the date of publication under the DOI: https://doi.org/10.6084/m9.figshare.25562667.v1. DOIs are also listed in the key resources table.
- Any additional information required to reanalyze the data reported in this paper is available from the lead contact upon request. Please note that detailed notes, instructions and reasoning for the analysis steps are included in the commentary of the code for data analysis.

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AUTHOR CONTRIBUTIONS

Conceptualization, J.S., M.D., and G.D.; Methodology, J.S., M.D., and G.D.; Software, J.S.; Investigation, J.S., M.D., A.E., M.K.V., A.S.L., and A.P.; Resources, M.D., G.D., P.S.Z., and A.E.; Data Curation, J.S. and M.K.V.; Writing – Original Draft, J.S. and M.D.; Writing – Review and Editing, J.S., G.D., A.E., A.P., M.K.V., A.S.L., P.S.Z., and M.D.; Visualization, J.S.; Supervision, M.D., G.D., and P.S.Z.; Project Administration, M.D., J.S., A.P., and A.S.L.; Funding Acquisition; M.D. and G.D.

DECLARATION OF INTERESTS

The authors declare no competing interests.

STAR★METHODS

Detailed methods are provided in the online version of this paper and include the following:

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STAR★METHODS

KEY RESOURCES TABLE

REAGENT or RESOURCE	SOURCE	IDENTIFIER
Software and algorithms		
Original code generated by this study.	This study.	https://doi.org/10.6084/m9.figshare.25562667.v1
MATLAB	The MathWorks, Natick, MA, USA	RRID: SCR_001622; https://www.mathworks.com/
EEGLAB	Swartz Center for Computational Neuroscience, La Jolla, CA, USA	RRID: SCR_007292; https://sccn.ucsd.edu/eeglab/index.php
Other		
Original data.	This study.	https://doi.org/10.6084/m9.figshare.25562667.v1

EXPERIMENTAL MODEL AND STUDY PARTICIPANT DETAILS

The study included 75 participants with a cleft, born between 2005 and 2011. The participants were between 11 and 18 years of age (adolescents). There were 36 male and 39 female participants, all were Slovenian residents and of Caucasian descent. We obtained the data on place of residence (three categories; either city area, suburban or small town area or rural area), education of the parents (according to the Slovenian Qualifications Framework,[51] ranging from 1 to 10, 1 = completed primary education and 10 = PhD) and their employment (number of employed parents) as indicators of socio-economic status, but large differences in the socio-economic status were not expected, as Slovenia has very high socio-economic equality (Gini index of 23.4).[52] Gender-based analyses were carried out.

All participants were treated at the Department of Maxillofacial and Oral Surgery of the University Medical Center Ljubljana, and were classified into three categories according to their diagnosis: the cleft palate only (CP) group, the cleft lip and palate (CLP) and cleft lip or lip and alveolus, but not palate group (CLA). All those willing to participate and not fitting any exclusion criteria were included in the study. The study was approved by the Medical Ethics Committee of Slovenia, Application No. 0120–365/2022/3, on 1.9.2022. The selection of participants is depicted in STAR Table A and presents the diagnosis, gender, socio-economic and age data.

STAR Table A. Summary of categorical variables and socio-demographic variables gathered on young people with clefts.

Variable	Categories	N	Missing (of 75)
Diagnosis	Cleft lip and palate (CLP)	30	5
	Cleft lip (CP)	26	
	Cleft lip and alveolus (CLA)	14	
Gender	Male	36	0
	Female	39	
Place of residence	City	21	2
	Suburban or small town	29	
	Rural	23	
Sum of employed parents	1	10	12
	2	53	
	Median	**Mean**	**Missing (of 75)**
Age (years)	14.721	14.703	8
Parents' education	6	5.786	12

In bold text are the names of columns or the name of the variable shown in the table.

METHOD DETAILS

The study was designed as a cross-sectional, observational, analytical study. The aim was to explain the differences in satisfaction with appearance using a multidimensional approach, combining the diagnosis of cleft, demographic characteristics, psychological characteristics and neurophysiological characteristics of participants. While the effects of different variables were compared across the CLP, CLA and CP groups, no control group was used. This is because the rationale for the study is to determine how satisfaction with appearance varies in young people

with clefts, and not in the general population, neither were we interested in the differences with the general population. A single participant represented a statistical (and experimental) unit.

A sample of 75 young people with a cleft was obtained. In the main model, the data of 40 participants was used, as 44 had complete data enabling regression analysis and four of these were outliers. In the first exploratory model, the data of 55 participants was used. In the second exploratory model, the data of 41. The third exploratory model (univariate frontal alpha asymmetry data) included the data of 47 participants. The sample consisted of all subjects willing to participate in the study, born between 2005 and 2011 and treated at the Department of Maxillofacial and Oral Surgery of the University Medical Center Ljubljana. *A-priori* power analysis (for the main model) showed that at least 77 participants were needed (with full data) to detect medium effect sizes ($f^2 = 0.15$) in our *a-priori* variables of interest (calculated using G*Power 3.1[53]). However, as less full data was available, we were only able to detect effect sizes of $f^2 = 0.27$ or greater for our variables of interest and $f^2 = 0.43$ or greater for all the other variables in the model (those to which the Bonferroni correction was applied – see the "quantification and statistical analysis" section).

The exclusion criteria were: incomplete medical documentation; the presence of a cleft due to syndromic disorders; chronic diseases and mental disorders; and head trauma or other medical conditions that could affect the results of the study. The inclusion criteria were consent, having an orofacial cleft and being born between 2005 and 2011. These criteria were established *a-priori*. No participants were excluded post-hoc and full data of all 75 participants is provided. No random selection occurred. To minimize masking effects of confounders, the multidimensional analysis approach was adopted. No blinding was utilized in the protocol of this study, as no treatment was applied.

The data was collected between 26.8.2022 and 15.11.2023. First, the medical documentation of all the children born with a cleft between 2005 and 2011 was screened, aiming to determine who fitted the study exclusion and inclusion criteria and to acquire data on the place of residence, age, and gender of the participants. The young people (and their parents) thus identified were approached and invited to participate in the study, either during their routine visits to the Dental Clinic of the University Medical Center of Ljubljana, or by telephone ahead of their routine visit. The protocol of the study was explained to them during this contact. Some potential participants expressed an interest in taking part in the study, but were unwilling to undergo the EEG section; they were offered participation by only filling in the questionnaires. The study protocol consisted of the administration of the questionnaires, which took 40-70 min to complete, followed by the acquisition of the resting-state EEG data; this took 20-40 min to complete. The parents' education and employment status were obtained by speaking to the parents by phone or during the routine visit. The parents' responses with regard to their education were first aligned with the Slovenian Qualifications Framework.[51] The framework has 10 levels (with 1 being the lowest), corresponding to education levels from completed primary education (level 1) to PhD (level 10). The mean of the education of the two parents (or one in the case of single-parent households) was then used as the final variable. The employment variable was obtained by summing the binary responses of the parent(s) to obtain the final variable.

At the end of the data collection, participants were asked about the tiredness and stress they currently experience. To make sure the young people understood the question, we explained stress as; "The unpleasant feeling you get when you have a lot of tests at school, have to do a lot of things, or have just had a fight with a friend. Rate how much stress you are feeling at the moment on a scale of zero to ten, where zero means that you are not stressed at all at the moment. Ten means that you feel you find it difficult to do everything that needs to be done, that you are very anxious about a current life situation (e.g., you are very distressed because you have had a fight with your friend)." As with stress, we asked children and young people with a cleft to rate their current level of tiredness on a scale of 0–10. We explained this; "A rating of zero means you feel like you have just woken up to a new day, full of energy and enthusiasm for new activities. You don't feel sleepy at all and you are well rested. A rating of ten means you feel like you can hardly stand up, and you can't wait to go to bed. You'd even rather skip dinner or lunch, meaning you feel completely exhausted, as if you've had a really hard day."

The four standardized questionnaires with high to very high reliability (see below) were administered in paper form.

(1) The Slovenian adaptation of the Inventory of Child Individual Differences (ICID, Slovenian abbreviation VMR-OM)[54] is a self-report measure of the personality traits that are perceived as most salient in young people. The questionnaire consists of 108 items and is scored on a 7-point Likert-type scale. The 15 traits are grouped into 4 superordinate personality traits: a) extraversion, pertaining to the level of activity, open-mindedness, openness to experience, positive emotionality and sociability; b) conscientiousness, pertaining to achievement orientation, agreeableness, openness, intelligence and organization; c) non-agreeableness, pertaining to antagonism, negative emotionality and strong will; and d) neuroticism, pertaining to fearfulness and social timidity. Cronbach's alpha was 0.809.

(2) The Slovenian adaptation of the Spanish original Questionnaire for the Evaluation of Interpersonal Difficulties in Adolescence (Spanish abbreviation CEDIA, Slovenian abbreviation VMTM),[55] is a 36-item self-report questionnaire consisting of 5 subscales: a) assertiveness, b) relations with the opposite sex, c) public speaking, d) relations with family, and e) relations with friends. The scale assesses the level of presence of interpersonal difficulties in a wide range of interpersonal relationships and social situations with people of different ages, genders, levels of authority and confidentiality in different settings (e.g., family, school, friends, gender relations) and situations (e.g., on the street, in the shop and other public places). Cronbach's alpha was 0.921.

(3) ABAS-3 is the Slovenian adaptation of the Adaptive Behavior Assessment System, Third Edition.[56] ABAS-3 helps us to identify problems in different areas of adaptive behavior. The system assesses adaptive behavior at three levels: the total adaptive behavior score, domains, and subdomains. The three domains and 11 subdomains consist of: conceptual (communication, functional educational skills, self-direction), social (interpersonal relationships, leisure) and practical (behavior outside the home, life at home and in kindergarten/school, health and safety, caring for oneself, work). Cronbach's alpha was 0.979.

(4) The CHASQ (Cleft Hearing, Appearance and Speech Questionnaire) is the Slovenian translation of the Appearance Satisfaction Questionnaire for People with Cleft Lip and/or Palate.[57] It consists of 15 items and two sub-scales; the first assesses category 1 characteristics, which rate satisfaction with the areas typically affected by a cleft. It is comprised of nine items and includes satisfaction with one's face, overall image, side profile, good looks, nose, lips, teeth, speech and a self-assessment of 'how noticeable my cleft is to other people.' The second, category 2 characteristics, cover self-assessment of satisfaction with areas not normally affected by a cleft – the chin, cheeks, hair, ears, eyes and hearing. The CHASQ gives a combined assessment of satisfaction with appearance, which is the sum of the two category scores and separate scores for each category. Cronbach's alpha was 0.944.

The results of the questionnaire data were obtained by inputting the data from the paper form questionnaires into custom scripts for evaluation, implemented in Microsoft Excel (The Microsoft Corporation, Redmond, WA, USA), according to the publisher's instructions.[54–57] The data was then collated into one file and imported to MATLAB 2023b (The MathWorks, Natick, MA, USA).

EEG data was prepared using the EEGLAB add-on for the MATLAB programming language.[58] Seven minutes of eyes open and 7 min of eyes closed resting state data was recorded for each participant. During EEG data acquisition, particular attention was paid to controlling environmental factors, to ensure equal conditions for all subjects. The data collection was carried out in a room at the Dental Clinic reserved for this purpose. Data on the brain activity of the subjects was acquired using the EEG device g.Nautilus (Guger Technologies, Graz, Austria), a system of 32 active wet EEG electrodes, mounted according to the 5/10 EEG electrode placement system. The sampling rate was set to 500 Hz, and a 48–52 Hz online notch filter was used. The EEG data was pre-processed by.

(1) The data was manually inspected for device malfunction data corruption or major environmental artifacts. No data was removed at this step.
(2) Filtering the data between 2 and 75 Hz, using two different filters (both finite impulse response filters, with a Hamming window, stopband attenuation of −53 dB and maximum passband deviation of 0.22%).
(3) First the highpass at 2 Hz, with a 2 Hz transition bandwidth and hence filter order of 414.
(4) Second the lowpass at 75 Hz, with a 10 Hz transition bandwidth and filter order of 166.
(5) Bad channels were automatically removed, using the criteria of flatline of more than 20 s, correlation of less than 0.8 with neighboring channels (as per electrode locations) and more than 5 SD of high frequency (above 30 Hz) power in comparison to all other channels.
(6) Independent component analysis (ICA) using the infomax ICA algorithm of Bell and Sejnowski[59] with the natural gradient feature of Amari, Cichocki, and Yang[60] and the extended ICA algorithm of Lee, Girolami, and Sejnowski.[61]
(7) We carried out a spherical spline interpolation of missing or removed electrode data, according to the procedure described in Ferree.[62]
(8) Last, a current source density transformation[63] was carried out, with the parameters λ = 0.00001, m = 3 and a uniform head radius = 10 cm for all participants.

After pre-processing the data, we computed the Welch's periodogram for all the EEG datasets, with a 2-s-wide Hann window with 50% overlap. Next, we extracted the power spectral density of the alpha frequency band (8–13 Hz) of each electrode and each EEG dataset. These power spectral densities were then used to calculate the frontal alpha asymmetry, using the following equation $\frac{\sum_j^{i(A_{ij}-B_{ij})}}{n}$, where 'i' is the selected frequency band; 'j' is the selected electrode location; 'A' is the decadic logarithm of the electrode power on the right side of the scalp; 'B' is the decadic logarithm of the electrode power on the left side; and 'n' is the number of electrodes included in the calculation. The electrodes included were those positioned over the frontal cortex, the AFp6, AFF10 h, F4, FT8 and FCC6 h on the right and the AFp5, AFF9 h, F3, FT7 and FCC5 h on the left. The obtained frontal alpha asymmetry values were then summed between the eyes open and eyes closed conditions. The EEG data pre-processing was carried out using MATLAB and its plug-in EEGLAB (v. 2023.1).[58]

QUANTIFICATION AND STATISTICAL ANALYSIS

We designed a linear regression model, aiming to determine how neurophysiological, psychological and demographic characteristics of young people with clefts associate with their satisfaction with appearance. Linear multiple regression modeling was carried out, including all viable variables, to allow the actual effect of each to be isolated. STAR Table A (see previous section) and STAR Table B show the summary of the categorical and numeric variables gathered.

STAR Table B. Summary of numeric variables gathered on young people with a cleft

		Median	Mean	Missing (of 75)
	Age (years)	14.721	14.703	8
The Inventory of Child Individual Differences (VMR-OM)	Conscientiousness	13.900	13.873	1
	Non-agreeableness	10.278	10.498	1
	Neuroticism	7.226	7.158	1

(*Continued on next page*)

Continued

		Median	Mean	Missing (of 75)
	Extraversion	23.495	23.276	1
Cleft Hearing, Speech and Appearance Questionnaire (CHASQ)	Category 1 characteristics	59.000	58.048	0
	Category 2 characteristics	53.000	49.080	0
	Entire score CHASQ	112.000	107.128	0
Interpersonal Difficulties in Adolescence Questionnaire (VMTM)	Relations with family	1.500	2.203	1
	Assertiveness	21.000	22.264	1
	Relations with the opposite sex	16.000	15.082	2
	Relations with friends	1.000	1.509	1
	Public speaking	4.000	5.601	1
	Entire score VMTM	52.000	46.873	2
Adaptive Behavior Assessment System (ABAS)	Conceptual facet	26.000	25.056	3
	Social facet	16.000	16.361	3
	Practical facet	32.000	32.333	3
	General score	73.000	73.750	3
	Frontal alpha asymmetry	−0.004	−0.003	21
	Tiredness	5.000	4.726	44
	Stress	2.000	1.887	44
	Parents' education	6.000	5.786	12

In bold text are the questionnaires for which the data was collected or single measures investigated (e.g., age, stress etc.).

All the statistical analyses were carried out in MATLAB, with the analysis scripts provided at the DOI under the subtitle "data and code availability" of this methods section. Before carrying out the planned analysis, we verified that the assumptions of the linear regression fitted our data. Diagnosis, gender, parents' employment and place of residence were included as categorical variables. Age, interpersonal difficulties, personality characteristics (extraversion, neuroticism, non-agreeableness and conscientiousness), adaptive behavior, frontal alpha asymmetry and education of the parents were included as numerical, continuous variables. The score of the CHASQ was used as the outcome variable for the model.

The assumption testing pipeline was.

(1) Verify the linearity of bivariate association between each explanatory variable and the outcome by scatter plotting.
(2) Calculating the Pearson correlation matrix of all variables to be able to better decide which variables to keep in the model during collinearity diagnostics.
(3) Diagnosing collinearity – first by examining the variance inflation factors of all variables (variance inflation factor must be <10) and then by using table plots; evaluating variance decomposition proportions by condition indices (variance decomposition proportion of two variables over 0.5 was considered to be problematic, but clustered proportions of over 0.3, that is multiple collinearities in more than one condition index was also considered to be problematic).
(4) Adaptive behavior scores were excluded due to severe or moderate collinearity among themselves and with other variables.
(5) Extraversion was eliminated due to collinearity with conscientiousness.
(6) The Durbin-Watson test was then ran on the initial linear model (DW = 1.917, p = 0.716).
(7) Histogram and Q-Q plots of the residuals were examined.
(8) The Breusch-Pagan test with the Koenker's modification was ran to verify the results of the histogram and Q-Q plots (p = 0.53).
(9) Outliers were identified using leverage and Cook's distance. Data points were considered outliers if their Cook's distances exceeded 3x of the mean of the Cooks' distance of all datapoints. For the leverage values criterion, the cutoff value was set as the $2 \times \frac{n_{coefficients}}{n_{obesrvations}}$. Four datapoints were excluded due to Cook's distance, and none due to leverage.
(10) The final model was then fitted.

All results were Bonferroni corrected for the number of unplanned comparisons, but otherwise the threshold of significance of $\alpha = 0.05$ was adopted.

Four exploratory analyses were also conducted, as surprising results were obtained with the original model. These were.

(1) A linear regression model on an extended number of subjects, excluding frontal alpha asymmetry (as this allowed the extension of the sample)
(2) As some differences in conclusions were arrived at as a result of the first exploratory model, a second one was fitted, to the data from model one, but without frontal alpha asymmetry
(3) As this once again produced differences in the results the third linear model was fitted – a univariate model between frontal alpha asymmetry and CHASQ scores
(4) In the end, five one-way ANOVA were carried out to verify whether the type of cleft significantly affected the significant variables in the models and the CHASQ score.

Exploratory models 1–3 underwent the same statistical pipeline as model 1. The normality of the distribution (Shapiro-Wilk's test of normality - violated in most cases) and equality of variance were tested for in the ANOVA. Welch's ANOVA was used where assumptions were violated. The results of the ANOVA were Bonferroni corrected for the number of ANOVA carried out. Post-hoc t or Mann-Whitney U (depending on the assumptions being violated or not) tests were likewise Bonferroni corrected.