DOES CLEFT REPAIR SURGERY RESTORE NORMAL VISUAL AND NEURAL RESPONSES TO INFANT FACES?

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Abstract

DOES CLEFT REPAIR SURGERY RESTORE NORMAL VISUAL AND NEURAL RESPONSES TO INFANT FACES?

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Infant faces readily capture our attention and elicit enhanced neural processing, likely due to their evolutionary importance in facilitating bonds with caregivers. Infant facial malformations are associated with a lower degree of parental investment and have been shown to negatively impact early infant-caregiver interactions. Cleft lip or cleft palate is a common facial malformation, estimated to affect 1 in 700 live births worldwide, that is associated with altered visual and neural processing as compared to normal infant faces. Importantly, it is not yet known how craniofacial repair surgery impacts responses to these faces. The current study uses eye tracking and electroencephalography (EEG) to investigate alterations in how adults process infant faces with cleft lip/palate following surgical repair. Results indicated that infants were rated as much cuter after craniofacial repair. Additionally, infant faces before surgery drew attention quicker and held attention longer at the mouth as compared to infants who underwent repair surgery, suggesting more normal visual processing for these faces after craniofacial repair. Findings also revealed supporting evidence of the restorative function of repair surgery for both the P2 and N170 components, but not the LPP, indicating that craniofacial repair surgery may restore more normative perceptual processing of infant faces, but not affective processing. As these differences may contribute to a number of important developmental aspects (e.g., joint attention) and may play a key role in the previously observed difficulties in caregiver-infant interactions, this study provides an important first step in determining the effectiveness of surgical interventions on the underlying neural mechanisms of infant face processing.

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Introduction

Relatively recent empirical evidence suggests that infant faces rapidly capture adults' attention (Brosch et al., 2007; Cardenas et al., 2013; Hodsoll et al., 2010) and elicit enhanced neural processing in comparison to adult faces (Kringelbach et al., 2008). It is suggested that this is likely due to evolutionarily important infant facial cues which may act as a relevant source of information for parental investment (reviewed in Bugental et al., 2010; Franklin & Volk, 2018). Infant facial cues have been noted to be key for multiple aspects of human communication including interpersonal interactions, maternal responsiveness, and perceived cuteness (reviewed in Lewis et al., 2017; Murray et al., 2008; Parsons et al., 2017).

Infant Cues & Health

Historically, research suggesting that infant faces are unique in terms of neural processing has been conducted using hegemonic standards, including exclusively normal, healthy infant stimuli. However, there is strong anthropological evidence noting that health-related infant facial cues influence caretaking behaviors (e.g., Hrdy, 1999; Volk et al., 2005). Among modern societies, cues of low health from infants remain associated with a lower degree of parental attention and care (Bugental et al., 2010; Coffey, 2006; Field & Vega-Lahr, 1984; Montirosso et al., 2012; Thamilselvan et al., 2015), and prenatal detection of fetal abnormalities can lead to increased rates of termination (see Hsieh et al., 2013). For example, Field & Vega-Lahr (1984) examined early face-to-face interactions between infants with and without cleft lip/palate and their mothers to investigate the potential that facial malformations impair parental investment. Infants with cleft lip/palate and their mothers both engaged in less smiling and vocalizing compared to healthy infant dyads, and mothers of cleft lip/palate infants showed overall fewer intimate behaviors than healthy infant mothers (Field & Vega-Lahr, 1984). Related data indicates that one of the most common historical and cross-cultural reasons for infant abandonment was cues related to poor health (e.g., Hrdy, 1999), with one study revealing that nearly 70% of abandoned children carried an appearance flaw that was neither life-threatening nor impactful for intellectual development (Weiss, 1994 as cited in Yamamoto et al., 2009).

Cleft Lip/Palate

One of the most commonly occurring facial abnormalities is cleft lip and/or palate, which consists of a facial malformation that affects the orofacial and sometimes nasal areas across faces. Cleft lip/palate has been estimated to affect 1 in 700 live births worldwide (Mossey & Catilla, 2003; Mossey et al., 2009), leaving affected children to experience a range of long-term adverse outcomes. Difficulties in early caregiver interactions have been explored in previous research, revealing that infants with more severe facial malformations receive less socially intimate behaviors as compared to infants with minor facial disruptions (Montirosso et al., 2012; Murray et al., 2008, 2018; Rayson et al., 2017; Speltz et al., 2000). Further evidence shows that not only are infant-caregiver interactions more difficult, but the reductions in nurturing behaviors directed towards infants with cleft lip/palate result in behavioral, socioemotional, and cognitive impairments (De Pascalis et al., 2017; Field & Vega-Lahr, 1984; Hardin-Jones & Chapman, 2011; Murray et al., 2008; Parsons et al., 2013; Rayson et al., 2017; Richman

& Eliason, 1982). For example, Kapp-Simon & Krueckeberg (2000) investigated the mental development of infants and toddlers with and without cleft lip/palate, and found that 45% of affected infants were declared as developmentally delayed after being tested on their cognitive function. These findings indicated that delays in acquisition of perceptual–motor skills and cognitive performance for infants with cleft lip/palate was greater than expected during their first year of life (Kapp-Simon & Krueckeberg, 2000).

Perceived Cuteness

Lorenz (1943) proposed that a specific set of physical characteristics act as a releasing mechanism for innate caretaking behaviors, including features such as that of a large head, round face, big eyes, small nose, and chubby cheeks. This effect, referred to as “kindchenschema”, is typically modeled in infant faces and is what adults typically use to determine whether an infant is perceived as “cute”. Numerous laboratory-based studies have shown evidence that infants with cleft lip/palate elicit different perceptual responses in adults than their healthy counterparts. Such studies have indicated that the presence of cleft lip/palate significantly reduces perceived cuteness of infant faces (Goodacre et al., 2004; Huffmeijer et al., 2018, 2020; Lewis et al., 2017; Parsons et al., 2011, 2013; Quast et al., 2018; Rayson et al., 2017; Yamamoto et al., 2009). As the perceived cuteness of infants is altered for infants with cleft lip/palate, disruptions to the typical release of caretaking behaviors may also put affected children at risk (Glocker et al., 2009; Kringelbach et al., 2016). The notable effects of cleft lip/palate on perceived cuteness have even been extended outside human faces, with domesticated animals being judged less cute when affected by a facial malformation (Parsons et al., 2011). Furthermore, studies using a keypress task have demonstrated that adults will exert effort to shorten their exposure to cleft lip/palate infant faces (Parsons et al., 2011; Yamamoto et al., 2009). Research exploring how the presence of cleft lip/palate impacts perceptual responses to infants is crucial to better understanding the influences it may have on later behavioral responses.

Further, suggesting that cleft lip/palate may disrupt responses associated with caretaking behavior (Kringelbach et al., 2008, 2016), reduced neural activity in a key reward-related region of the parental brain, the orbitofrontal cortex (OFC), has been observed when adults viewed cleft lip/palate infants in comparison to healthy infants (Parsons et al., 2011, 2013). In addition, adults show weaker activity in the fusiform face area (FFA), a portion of the brain sensitive to the processing of faces, when viewing infant faces with cleft lip/palate compared to non-affected controls (Parsons et al., 2011, 2013). The diminished neural responses (Huffmeijer et al., 2018; Parsons et al., 2011) could explain why adults are less motivated to view affected infants and why adults tend to rate them as less cute in comparison to their healthy counterparts (Rayson et al., 2017; Yamamoto et al., 2009).

Eye Gaze

Humans hold a strong bias toward looking at the eyes when viewing a face, which indicates that the eyes play a critical role in capturing attention and directing social attention (reviewed in Birmingham & Kingstone, 2009; Langton et al., 2000). Furthermore, direct, joint attention with infants and their caregivers is crucial to the development of later key cognitive and social processes (Britton et al., 2001; Rayson et al., 2017). Mutual gaze between parents and infants has been associated with increased engagement during face-to-face interactions, reinforcement of early social expressiveness in infants, and has been linked to the quality of their relationship (Britton et al., 2001; De Pascalis et al., 2017; Hains & Muir, 1996; Rayson et al., 2017). It is possible that the presence of cleft lip/palate may cause disturbances in early parent-infant interactions due to drawing attention away from the eyes, which has been linked to an array of unfavorable outcomes such as the reduction of maternal responsiveness and decreased sensitivity to infant communicative cues (De Pascalis et al., 2017; Field & Vega-Lahr, 1984; Montirosso et al., 2012; Murray et al., 2008; Rayson et al., 2017). Studies using eye tracking revealed that adults and mothers fixated on the mouth region of the faces of infants with cleft lip/palate at the expense of fixation on the eyes in comparison to healthy infants (De Pascalis et al., 2017; Hahn et al., 2023; Rayson et al., 2017), and that the severity of cleft lip/palate predicted this fixation bias (Rayson et al., 2017).

Rayson and colleagues (2017) used eye tracking to show a positive correlation between cuteness ratings and time spent fixated on the eyes. Findings indicated that the cuter an infant was rated, the greater the proportion of time participants spent fixating on the eyes compared with the mouth, with cleft lip/palate infants being rated less cute and receiving more time fixated on the mouth at the expense of the eyes. They also found that participants fixated longer on the mouth of cleft lip/palate faces that were rated as less cute. Similarly, work in our lab that motivated the current study found that adult visual attention was rapidly captured by the mouth region for infants with cleft lip/palate, at the expense of the eyes (Hahn et al., 2023). De Pascalis et al. (2017) reported that the presence of cleft lip/palate fostered increased maternal gaze to the mouth, at the expense of the eyes, and that gaze towards infant faces, overall, was reduced for affected infants. Findings also indicated that reduced visual attention to infants with cleft lip/palate occurred during the critical developmental window where social expressiveness emerges, and that this altered maternal gaze may risk maternal fostering of infant social communicative cues that would normally occur for non-affected infants (De Pascalis et al., 2017). Together, these findings (De Pascalis et al., 2017; Hahn et al., 2023; Rayson et al., 2017) suggest a relationship between perceptions of cuteness and gaze patterns, which can in turn impact key reward regions of the brain associated with maternal responsiveness (Parsons et al., 2013). Furthermore, it has been demonstrated that changes in gaze patterns, such as those observed in the presence of cleft lip/palate, can have dual impacts of reducing mother-infant bonding (Murray et al., 2008) and, at the same time, increasing the negative feelings of the mother (Stone & Potton, 2019).

Neural Processing

Previous work investigating neural responses to infant faces has also provided evidence of disruption to normative processing as a result of cleft lip/palate (Hahn et al., 2023; Huffmeijer et al., 2018, 2020; Parsons et al., 2013). Studies using electroencephalography (EEG) to investigate facial neural processing typically explore the P1, P2, N170, and LPP ERP components (Bentin et al., 1996; Eimer, 2000; Hahn et al., 2016, 2023; Huffmeijer et al., 2018; Rossion et al., 2003).

P1

The P1 is an early positive event-related-potential (ERP) component, occurring at approximately 100 ms after stimulus onset. It is observed at posterior electrode sites, and is thought to reflect early perceptual processing of low-level stimulus features such as contrast (Itier & Taylor, 2004) and spatial frequency (Halit et al., 2000). The P1 is also believed to be sensitive to disruptions or alterations in the configuration of facial features (Halit et al., 2000; Linkenkaer-Hansen, 1998).

N170

The N170 is an early, negative ERP component, which peaks at approximately 170 ms after stimulus onset. This face-sensitive component is the most widely used in face perception research, and is measured over occipitotemporal areas of the scalp (Eimer, 2000, 2011). N170 amplitude is thought to represent the structural encoding of faces associated with configural processing (Bentin et al., 1996; Eimer, 2000) and, more recently, to both positive and negative aspects of parental behavior (Bernard et al., 2015; Rodrigo et al., 2011; Rutherford et al., 2017). Configural processing of faces includes the interpretation of spatial relationships between facial features and considers the holistic arrangement of such features, rather than encoding each individual element, such as while engaging in featural processing (Bentin et al., 1996; Eimer, 2000). Disruptions to configural processing have been investigated by studies using both upright and inverted facial stimuli, as the upside-down presentation of a face can be more difficult to encode due to altered and unfamiliar facial configurations (Eimer, 2000, Yin, 1969). Specifically, larger, more negative N170 amplitudes are typically observed for inverted faces (Bentin et al., 1996; Eimer 2000), and can act as a control to compare with upright stimuli to further investigate the underlying mechanisms of facial configural processing.

P2

The P2 is a later, positive ERP component which occurs at approximately 200 ms post stimulus onset. It is observed at posterior electrode sites over brain areas which assist with visual encoding and are sensitive to facial configuration (Halit et al., 2000; Rossion et al., 2003; Schweinberger & Neumann, 2016). The P2 is thought to reflect encoding of second order spatial relations of faces, in particular, and is sensitive to the typicality of faces (e.g., Rossion et al., 2003; Schweinberger & Neumann, 2016). Specifically, Halit et al. (2000) observed increased P2 amplitudes when participants were presented with prototypical stimuli (i.e., typical spatial relationships between features) as compared to faces that were further away from prototypicality (i.e., 20% and 30% greater distance between facial features than normal). Alterations of P2 responses for atypical faces suggest that when continually exposed to a series of stimuli, top-down prediction of the next stimulus starts to emerge based on a prototype, and therefore, could explain why this component is sensitive to deviations from prototypicality (Halit et al., 2000).

LPP

The late positive potential (LPP) is a positive ERP component which begins at approximately 400 ms after stimulus onset. It is measured over central midline electrode sites on the scalp, and is thought to be modulated by the motivational relevance of stimuli (Schupp et al., 2000, 2004). LPP responses are typically elevated after the presentation of emotionally salient and arousing stimuli (Hajcak et al., 2009; Liu et al., 2012; Schupp et al., 2000, 2004), although the valence (i.e., positive or negative affect) cannot be explained solely by the intensity of the amplitude. For example, Schupp et al. (2004) found larger LPP responses to threatening faces as compared to faces with neutral and happy facial expressions, suggesting that emotional arousal is elevated considering contextual relevance of the stimuli. Overall, the LPP is a useful tool which provides insights into the allocation of cognitive mechanisms associated with the emotional processing of arousing stimuli.

ERP Responses to Cleft Lip/Palate

Two research groups (Hahn et al., 2023; Huffmeijer et al., 2018) have compared ERPs in responses to non-affected and cleft lip/palate infants. No significant differences were found for P1 amplitude in either study, revealing that cleft lip/palate has little to no effect on early lower level visual processing. However, both research teams saw reduced activity for P2 components in response to infants with cleft lip/palate as compared to normal infants. Attenuation of the P2 response for faces with cleft lip/palate suggests possible increased distance of these faces from facial typicality (Hahn et al., 2023; Halit et al., 2000; Schweinberger & Neumann, 2016), leading to the assumption that affected infants are processed as atypical.

Both groups also found a significant effect for N170 amplitude while adults viewed infant faces with cleft lip/palate, yet the direction of the effect observed was different (smaller, less negative amplitude for cleft lip/palate infants in comparison to healthy infants for Huffmeijer et al. (2018) and a larger, more negative amplitude for cleft lip/palate infants in comparison to healthy infants for Hahn et al. (2023). Further, findings from Hahn et al. (2023) suggest that the recruitment of additional processing mechanisms may take place while interpreting the faces of infants with cleft lip/palate. Sadeh & Yovel (2010) argue that configural disruptions bring facial stimuli closer to that of non-face objects, and therefore additional non-“face-selective” neural mechanisms (Yovel, 2016) must be engaged. One explanation for Huffmeijer et al.’s (2018) finding that N170 amplitudes were attenuated for cleft lip/palate infants could be due to the drawing away of attention from the eyes to the mouth, as is supported by Bentin et al.’s (1996) work finding that the N170 is greatest for the eyes as compared to other facial areas. Although in opposite directions, a direct effect of cleft lip/palate on early neural processing (i.e., N170) was observed by both teams, indicating that configural processing is impacted by the presence of facial malformations. However, the configural disruption of cleft lip/palate causing an increased N170 response is more in line with the previous work indicating that configural disruptions typically result in increased N170 responses (Bentin et al., 1996; Carbon et al., 2005; Eimer, 2000; Itier & Taylor, 2004).

Various configural conditions have indicated disruptions to normal N170 responses, supporting evidence for increased amplitudes in response to altered stimuli (Bentin et al., 1996; Carbon et al., 2005; Eimer, 2000; Itier & Taylor, 2004). First, Bentin et al. (1996) found a relationship between inverted faces and greater, more negative amplitudes when compared to upright faces. Similarly, Eimer (2000) also found that the impact on N170 amplitude from inversion expands even outside facial stimuli, showing that both inverted faces and pictures of houses stimulated an enhanced N170 response. These findings suggest that the inversion of stimuli disrupt typical configural processing, and in turn amplify N170 activity (Bentin et al., 1996; Eimer, 2000). In line with these studies, contrast reversed images of faces also have been demonstrated to elicit an increased N170 response, suggesting that with disruptions to facial configuration, the recruitment of additional mechanisms is required to interpret the stimuli properly (Itier & Taylor, 2004). Another study found enhanced N170 responses for thatcherized faces (i.e., the eye and mouth regions of the face are turned upside-down), further indicating that it is typical to observe larger N170 amplitudes in the presence of any configural disruption (Carbon et al., 2005). As cleft lip/palate significantly alters the typical facial configuration of infant faces, it is expected that the brain will process this as a configural disruption, and in turn elicit larger N170 responses as compared to the opposing stimuli.

Hahn and colleagues (2023) also investigated LPP as an additional ERP component which showed heightened responses to infants with cleft lip/palate, demonstrating increased emotional processing for affected infants. Together, despite differences in N170 responses, both Huffmeijer et al. (2018) and Hahn et al. (2023) have extended previous findings supporting the idea that cleft lip/palate alters early (NI70) and late (P2, LPP) neural responses to infant faces, likely due to the configural disruption cleft lip/palate causes.

Cleft Lip/Palate Repair Surgery

Although there is growing empirical evidence that cleft lip/palate disrupts visual and neural processing and affective behavior, it is not yet known how surgical repair impacts responses to these faces. Craniofacial interventions for cleft lip/palate focus on restoring both the normal function and aesthetic appearance of the orofacial and nasal areas of the face, including the upper lip, hard and soft palates, and nose (Campbell et al., 2010; Sandberg et al., 2002). Dramatic cosmetic changes come from craniofacial repair surgery, which is typically performed either during the neonatal period or roughly 3 to 4 months postpartum (Campbell et al., 2010; Parsons et al., 2013; Sandberg et al., 2002). Because 5,700 of the 4 million infants born each year have failed embryological development of lip and palate, there is a high demand for cleft lip/palate repair surgery (Posnick & Kinard, 2021; Sandberg et al., 2002). Few reports on the prevalence of craniofacial repair surgery for cleft lip/palate have been published worldwide, although, relatively recent research reports that 48% of infants with unilateral cleft lip/palate and 65% of infants with bilateral cleft lip/palate require repair surgery (Daskalogiannakis & Mehta, 2009).

Snowden et al. (2003) revealed that those receiving craniofacial repair surgery including that of cleft lip/palate result in an average cost of $11,350 per case of anomaly. Yet, the Centers for Disease Control and Prevention (CDC, 2007) also reported that infants with both cleft lip and palate have a mean hospital charge of $33,387, and that infants with cleft lip (with or without cleft palate) have a mean hospital charge of $15,387. Recent research from Boulet et al. (2009) further found that the mean and median costs for children with cleft lip/palate were approximately eight times greater than for non-affected children, which suggests a substantial economic burden for families with children who are born with the facial malformation. Considering these averages were calculated between ten and twenty years ago (Boulet et al., 2009; CDC, 2007; Snowden at al., 2003), the costs of neonatal hospital charges for affected infants have most likely increased dramatically. Whether craniofacial repair surgery may help to restore adults' visual and neural responses to those seen for typical infant faces is crucial to investigate, considering the frequency of both cleft lip/palate and repair surgeries performed, and the financial impact it may have on the families of affected infants.

Studies comparing the effects of neonatal versus later repair resulted in comparable outcomes of cuteness ratings, suggesting that regardless of timing, perceived attractiveness increases with craniofacial repair surgery (e.g., Goodacre et al., 2004; Murray et al., 2008). However, Murray and colleagues (2008) did find that infants who received later repair resulted in impaired cognitive functioning in comparison to infants who underwent repair during the neonatal period. These findings are noteworthy because their data further suggests that early difficulties in the interaction between mothers and affected infants mediate the cognitive impairment. Cleft lip/palate repair has been considered a safe intervention for affected infants at both the neonatal and later ages, and has been seen to have positive effects on parent-infant social bonding (Sandberg et al., 2002), feeding habits, and speech and physical development (Campbell et al., 2010; Sandberg et al., 2002). Continuing investigation of behavioral responses and visual and neural processing is of high importance to further develop a more profound understanding of how cleft lip/palate repair surgery affects parent-infant interactions and the social, cognitive, and behavioral development of affected infants.

The Current Study

The current study aims to determine whether cleft lip/palate repair surgery restores a more normal pattern of visual scanning and face processing responses for infants before and after craniofacial repair surgery. Based on previous findings (Hahn et al., 2023; Huffmeijer et al., 2018; Lewis et al., 2017; Rayson et al., 2017), it is expected that: (1) craniofacial repair surgery will increase perceived cuteness, (2) craniofacial repair surgery will restore more normative visual scanning of infant faces such that visual attention to the eyes will be prioritized for infant faces who underwent surgery, while visual attention to the mouth will be prioritized for infant faces before surgery, and (3) craniofacial repair surgery will impact both the early (N170) and late (P2, LPP) neural processing of infant faces. Specific hypotheses are outlined below:

Hypothesis 1. Cuteness Rating

It was predicted that craniofacial repair surgery will enhance cuteness, and thus infant faces will be rated as significantly cuter after surgery as compared to before surgery.

Hypothesis 2a. Time to First Fixation

It was predicted that an interaction between palate and face region (i.e., eyes and mouth regions of the face) will be observed for the time to first fixation variable. Time to first fixation at the eyes should be faster for infant faces after craniofacial repair surgery as compared to infant faces before surgery, while time to first fixation at the mouth should be faster for infant faces before surgery than infant faces after craniofacial repair. Simply, infants before surgery should draw faster attention to the mouth versus the eyes in comparison to infants after repair surgery, and infants who had craniofacial repair should draw faster attentional orientation to the eyes versus the mouth compared to infants before surgery.

Hypothesis 2b. Duration to First Fixation

It was predicted that an interaction between palate and face region will be observed for the duration of first fixation variable. Duration of first fixation at the eyes should be longer for infant faces after craniofacial repair surgery as compared to infant faces before surgery, while duration of first fixation at the mouth should be shorter for infant faces after surgery as compared to infant faces before surgery. Essentially, cleft lip/palate infants should hold the attentional orientation of participants longer at the mouth versus the eyes compared to infants who underwent craniofacial repair, and repaired infants should hold the attentional orientation of participants longer at the eyes versus the mouth compared to infants before surgery.

Hypothesis 3a. P1

Because previous work has not found an impact of cleft lip/palate at the P1 component, no significant effects or interactions were predicted here.

Hypothesis 3b. N170

Because the N170 reflects configural processing, a significant interaction between orientation and palate was predicted. Based on earlier work from our lab group (Hahn et al., 2023), it was predicted that N170 amplitude will be larger for infant faces before repair surgery (where facial configuration is more disrupted) than after repair surgery (where facial configuration is more normal). Because inversion disrupts face configuration, the impact of craniofacial repair surgery should be more apparent for upright than inverted faces.

Hypothesis 3c. P2

Because the P2 is impacted by the typicality of a face, a significant interaction between orientation (i.e., upright or inverted face presentation) and palate was predicted. It was expected that P2 amplitudes will be larger for infant faces after repair surgery as compared to before surgery in the upright condition. This is because the faces should have a more typical baby appearance following the cleft lip/palate repair surgery than they do before surgical intervention. Because we usually only see faces upright, inverted faces already deviate from face typicality. As such, the impact from repair surgery should be greater for the upright than inverted condition.

Hypothesis 3d. LPP

Based on our previous work (Hahn et al., 2023), no interaction between orientation and palate was predicted for the LPP, as emotionally relevant stimuli should elevate affective response regardless of orientation. A significant main effect of palate should be seen for LPP, with greater amplitude expected for faces before repair surgery as compared to after repair surgery. As the severity of facial malformation is greater for infants with cleft lip/palate than infants after undergoing repair surgery, a larger response for emotional salience should be expressed.

Methods

Participants

Twenty-six participants (17 female, 9 male) were recruited from the local Humboldt area by convenience sampling and word of mouth. Data loss occurred for four of the participants in total, resulting in a sample size of 24 participants for the cuteness rating task, 23 participants for the eye tracking measure, and 22 participants for the EEG portion of the study. Participants ranged in age from 19 to 59 years (*M* = 29.96, *SD* = 11.8). Six of them reported having children, and four participants reported working with children in some capacity (e.g., daycare; mean reported time per week spent around children = 3.86 hours). The sample was primarily right-handed (N = 21) and mostly Caucasian (N = 13). Most participants reported normal or corrected-to-normal vision (N = 23).

Stimuli

Full color photographs of 35 infants before and after craniofacial repair surgery were collected from an online search. These primarily came from before/after photographs displayed on plastic surgeons’ websites. To be eligible stimuli, infants were required to be facing the camera head on with open eyes and no other individual (i.e., adult hands holding the head, etc.) visible in the image. Efforts were made to ensure the before and after images selected were as similar as possible, although infants were consistently older in the after photo in comparison to the before (this difference was minimized to the best of our ability). All images were aligned on interpupillary distance and masked with a white background to remove any non-face information (done via

Webmorph; Debruine, 2018). The faces were presented in full color at a size of 600x800 pixels.

Procedure

For the duration of the study, participants were seated on a height-adjustable chair and instructed to minimize eye and body movements during the recording period. Head movement was limited via a chin rest (Tobii) located 85 cm from the computer screen (26” DELL monitor). Stimulus presentation and response recording was done using OpenSesame (Mathôt et al., 2012).

Cuteness Rating

Following the presentation of each infant face during the eye tracking portion of the study (see below), participants rated the cuteness of the face they had just seen on a scale from 1 (not cute) to 5 (very cute). The infant faces were presented in a fully randomized order, including both cleft lip/palate and repaired stimuli.

Eye Tracking

Eye movements were recorded using the GazePoint GP3 infrared eye tracker sampling at 60 Hz. The eye-tracker was placed below and slightly in front of the monitor, approximately 72 cm from the chin rest. Prior to beginning the study, a 9-point calibration was performed. Before each trial, a fixation cross appeared at the center of the screen and was immediately followed by an infant face, displayed for 5 seconds. After exposure to each image, participants rated the cuteness of the face.

EEG Acquisition

During EEG recording, participants were instructed to simply view the faces presented on the screen (i.e., a passive viewing task). Each of the 70 (35 before and 35 after repair surgery photos) infant faces were displayed twice in both an upright and inverted orientation, for a total of 280 trials. Each trial began with the presentation of a fixation cross at the center of the screen, followed by the presentation of a single face for 500 ms. Intertrial intervals varied randomly between 1000-1500 ms.

EEG was continuously recorded from 64 scalp sites, using BioSemi ActiveTwo Ag/AgCl electrodes and hardware (Biosemi, Amsterdam, The Netherlands). The electrodes were placed according to the 10-5 electrode system (Oostenveld & Praamstra, 2001), using a nylon electrode cap. EEG signals were amplified with a bandpass of DC-104 Hz by BioSemi ActiveTwo amplifiers, sampled at 512 Hz. Individual trial epochs with large artifacts (+- 500 uV) were removed before applying an independent component analysis (ICA) approach to artifact removal (Jung et al., 2000). The data was cleaned by removing any components that had both a less than a 10% chance of being labeled as originating in the brain and a greater than 60% chance of originating from the eye, heart, muscle, or power line noise. The cleaned trials were then averaged separately for each condition.

Data Processing

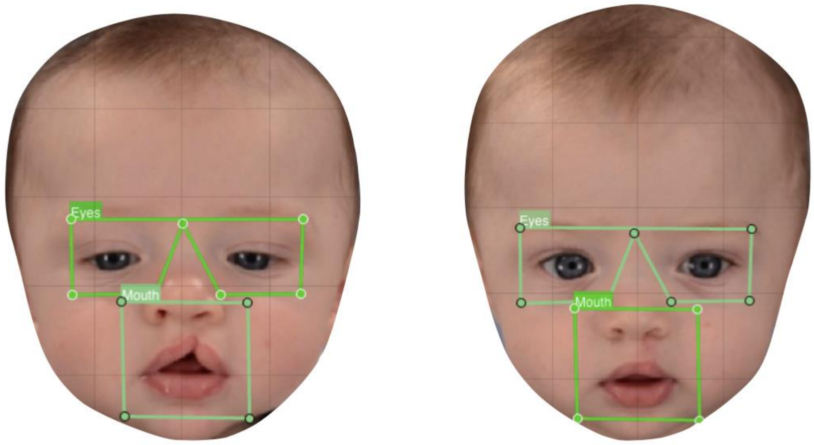
Eye Tracking

Two regions of interest (ROIs) were manually selected for each image. The eye ROI used encompassed both eyes and was rectangular with the exception of the bottom central border which follows the bridge of the nose near the nasion. The mouth ROI encompassed the mouth and the lower portion of the nose, extending to the tip of the nose. Although they differ in shape, the area covered by each of the two ROIs was similar (see Figure 1).

For each ROI, time to first fixation and duration of first fixation were calculated. Time to first fixation captures how quickly participants look at the mouth and eye region of the face and provides a quantitative measure of the power of the cleft lip/palate to draw attention. Duration of fixation reveals how long such regions of interest hold participants' attention.

Figure 1

Eye and Mouth Regions of Interest for Eye Tracking Measure



EEG

ERP components were quantified for each participant and condition by averaging the signal within a time and channel montage that captures the spatiotemporal properties of each ERP component. Time windows and channel montages were selected based on previous literature and confirmed by visual inspection of the global field power (for timing) and scalp maps (for montages) of the grand averaged data. Following our previous study on cleft lip/palate versus normal faces (Hahn et al., 2023), four “face perception” ERP components were analyzed: P1, N170, P2, LPP. The P1 was defined in a window between 100 and 150 ms and across a set of occipital electrodes that included PO3, POz, PO4, O1, Oz, O2 and Iz. The N170 was evaluated as between 155 and 215 ms and included electrodes P7 and P9 in the left hemisphere and P8 and P10 in the right hemisphere. The P2 was defined in a window from 220 to 270 ms in the same occipital electrodes as the P1. Finally, the LPP was evaluated as between 500-600 ms across a set of central parietal electrodes including CPz, Pz, P03, P0z, P4.

Results

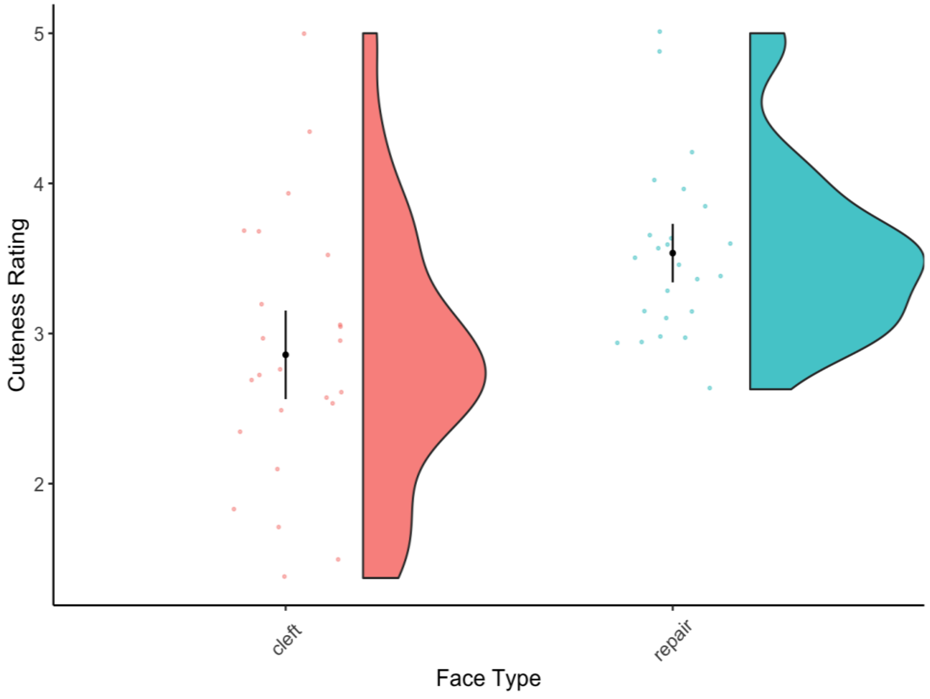
All data preprocessing and statistical analyses were conducted in Matlab and R, respectively.

Cuteness Rating

A paired samples t-test confirmed that infants were rated as significantly cuter after undergoing craniofacial repair surgery ($t(23) = -6.88$, $p < .001$, $d = -1.40$, see Figure 2). Infants with cleft lip/palate ($M = 2.86$, $SD = 0.88$) were judged as less cute in comparison to their repaired counterparts ($M = 3.54$, $SD = 0.58$).

Figure 2

Cuteness Ratings for Infants Before and After Craniofacial Repair Surgery



Eye Tracking

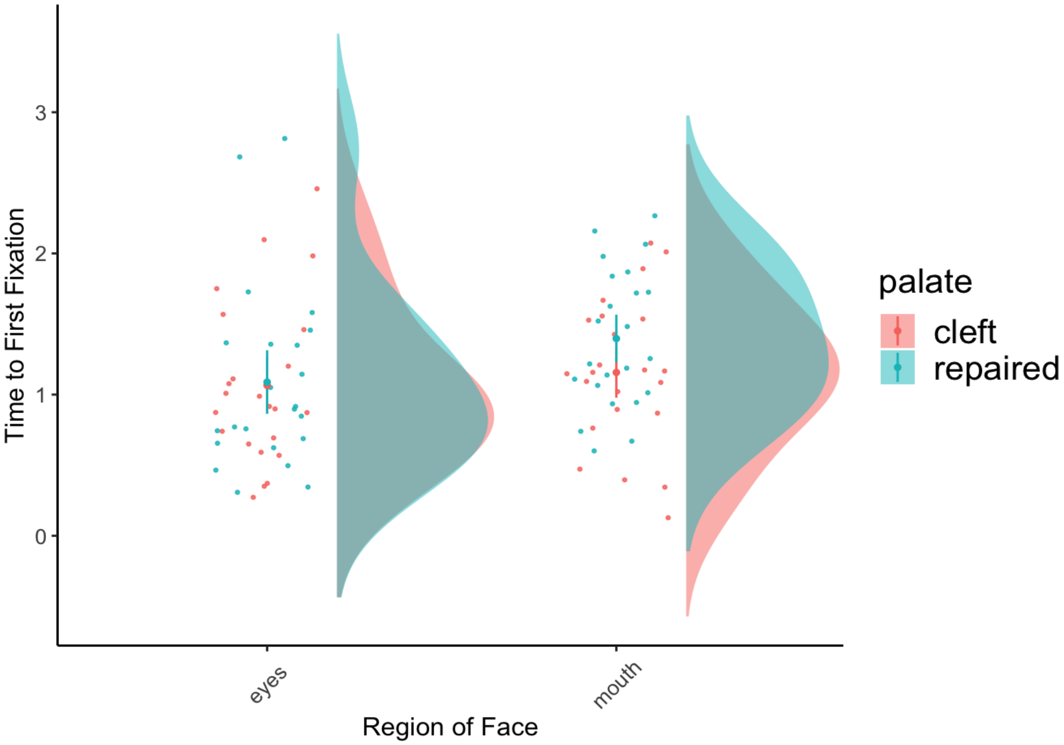
Time to first fixation and duration of first fixation were analyzed using two 2x2 repeated measures ANOVAs with ROI (eyes, mouth) and palate (cleft, repaired) as within-subject factors.

Time to First Fixation

A main effect for palate was observed (*F*(1, 22) = 7.63, *p* = .011, η2 = .014), but there was no main effect for ROI (*F*(1, 22) = 1.47, *p* = .239, η2 = .032). However, a significant interaction between palate and ROI (*F*(1, 22) = 4.71, *p* = .041, η2 = .009, see Figure 3) was found. Bonferroni corrected pairwise comparisons were used to further explain the interaction. Results showed that time to first fixation of the mouth region was significantly earlier for infants before repair surgery (*M* = 1.16, *SD* = 0.52) than after surgery (*M* = 1.39, *SD* = 0.49), indicating that infants before craniofacial repair draw attention to the mouth much more rapidly than infants after repair surgery, *t*(22) = -3.33, *p* = .003. This reveals a significant impact of craniofacial repair surgery on where adults fixate on infant faces who underwent repair. Conversely, time to first fixation of the eye region of the face showed no significant differences for infants after craniofacial repair surgery (*M* = 1.08, *SD* = 0.66) as compared to infants before surgery (*M* = 1.07, *SD* = 0.58), indicating that the prominent capturing of attention by the mouth had a stronger effect on the interaction as compared to the eyes, *t*(22) = -0.36, *p* = 0.725. Essentially, a more normal visual processing pattern took place for infants who underwent repair surgery, suggesting that the presence of the mouth was less capturing after surgery as compared to before repair.

Figure 3

Palate x ROI Interaction for Time to First Fixation



Duration of First Fixation

Results, again, revealed a main effect of palate (*F*(1, 22) = 20.87, *p* < .001, η2 = .037), but no effect of ROI (*F*(1, 22) = 1.30, *p* = .268, η2 = .013). However, a significant palate x ROI interaction (*F*(1, 22) = 9.05, *p* = .006, η2 = .013, see Figure 4) was observed. Follow up, Bonferroni corrected pairwise comparisons revealed that this interaction occurred because duration of first fixation on the mouth region of the face was significantly longer for infants before craniofacial repair surgery (*M* = 0.52, *SD* = 0.15) as compared to infants after surgery (*M* = 0.43, *SD* = 0.08), *t*(22) = 5.05, *p* < .001. Such results indicate that infants before craniofacial repair hold the attention of adults at the mouth region of the face significantly longer than infants after surgery, suggesting that craniofacial repair surgery helps to restore more normative visual patterns for infants who underwent repair surgery. No differences in duration of first fixation for the eye region of the face were found for infants before (*M* = 0.52, *SD* = 0.21) and after (*M* = 0.50, *SD* = 0.16) craniofacial repair, *t*(22) = 1.49, *p* = .150.

Figure 4

Palate x ROI Interaction for Duration of First Fixation

EEG

For the P1, N170, and P2 components, a 2x2x2 repeated measures ANOVA was used with orientation (upright, inverted), palate (cleft, repaired) and hemisphere (left, right), as within-subject factors. However, in line with our previous findings (Hahn et al., 2023), hemisphere showed no significant interactions, so the data was collapsed to create a 2x2 (orientation x palate) ANOVA for each component. As the LPP is measured centrally on the scalp and is not bilateral, a 2x2 repeated measures ANOVA was used with orientation (upright, inverted) and palate (cleft, repaired) as within-subject factors for this component.

P1

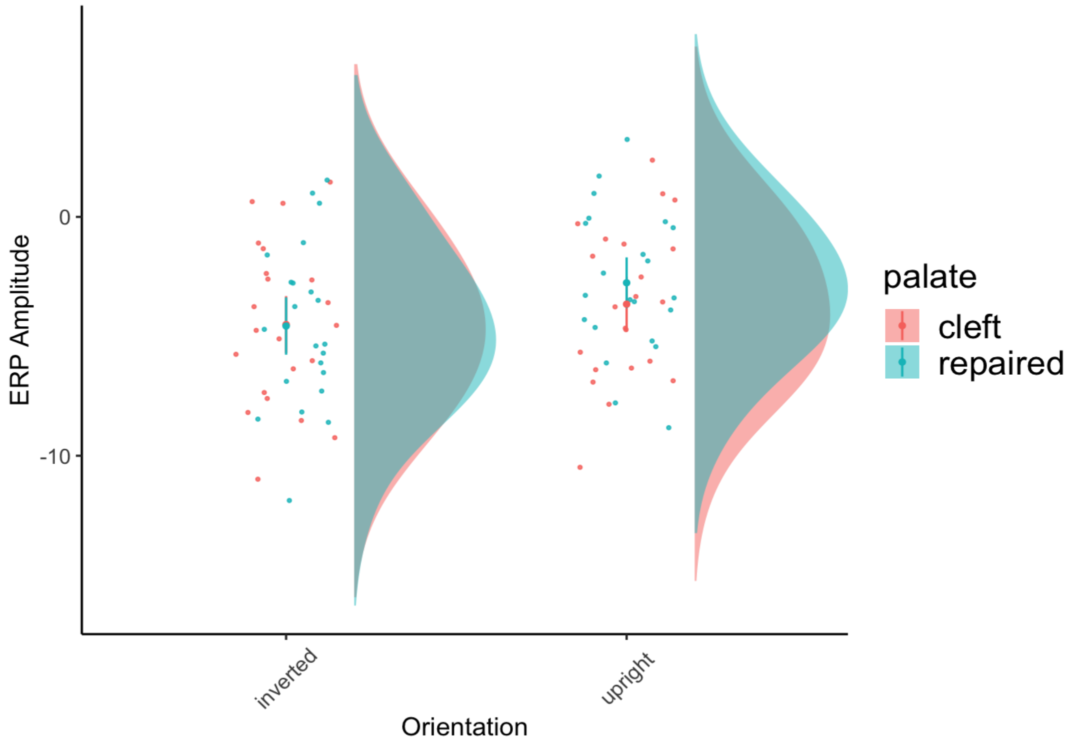
A main effect of orientation (*F*(1, 21) = 25.21, *p* < .001, η2 = .076) was found, with inverted faces (*M* = 4.01, *SD* = 2.28) evoking a larger P1 response compared to upright faces (*M* = 2.72, *SD* = 2.27). No other effects or interactions were significant (all p > .01).

N170

Results revealed main effects for both orientation (*F*(1, 21) = 41.06, *p* < .001, η2 = .041) and palate (*F*(1, 21) = 10.18, *p* = .004, η2 = .004), as well as a significant orientation x palate interaction (*F*(1, 21) = 15.76, *p* < .001, η2 = .006). Bonferroni corrected pairwise comparisons explained that this interaction resulted because infant faces before craniofacial repair surgery (*M* = -3.65, *SD* = 3.27) generated a larger N170 response versus infant faces after surgery (*M* = -2.76, *SD* = 3.03) when upright (*t*(21) = -4.24, *p* < .001), but not when inverted (*t*(21) = 0.46, *p* = .652, see Figure 6). N170 responses for inverted infant faces before surgery (*M* = -4.51, *SD* = 3.40) were approximately the same as inverted infant faces after surgery (*M* = -4.57, *SD* = 3.42). These findings suggest that recruitment of additional facial processing mechanisms may be required while viewing infants faces before craniofacial repair surgery, and that participants can rely more on configural processing when processing faces of infants after undergoing craniofacial repair.

Figure 5

Orientation x Palate Interaction for N170 Component

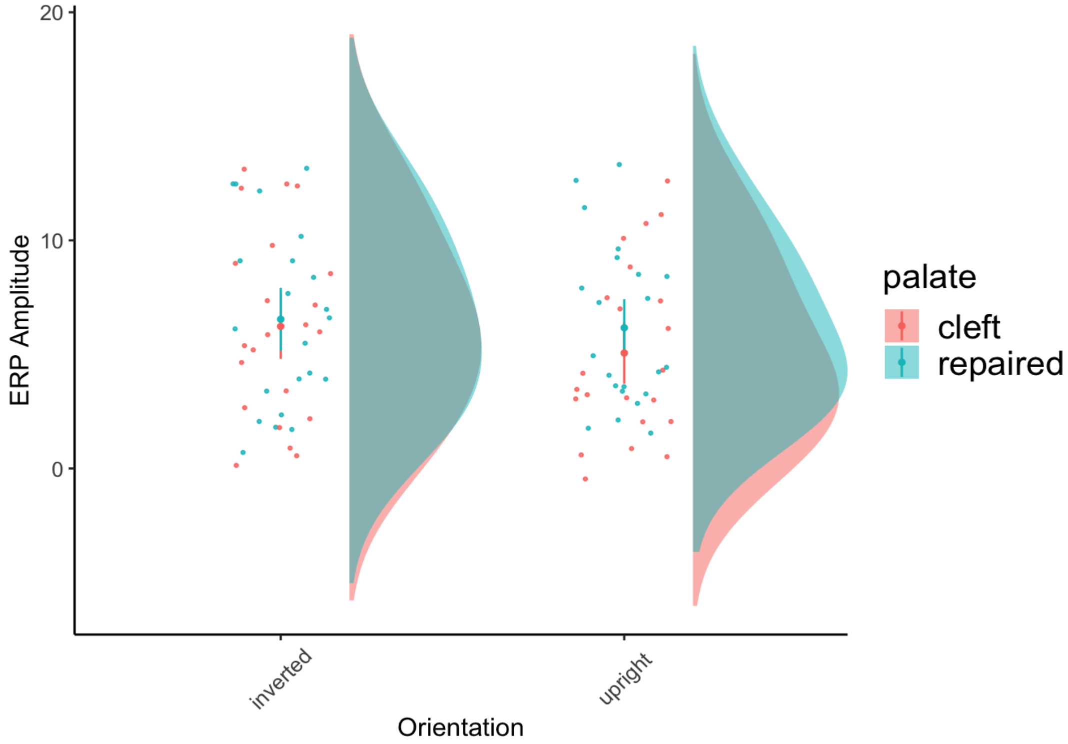


P2

Main effects were observed for both orientation (*F*(1, 21) = 8.75, *p* = .008, η2 = .010) and palate (*F*(1, 21) = 20.29, *p* < .001, η2 = .009). Furthermore, an interaction between orientation and palate was revealed for the P2 component, *F*(1, 21) = 12.85, *p* = .002, η2 = .002. A post hoc Bonferroni corrected pairwise comparison indicated that the interaction occurred because P2 responses were larger for infants after craniofacial repair surgery (*M* = 6.17, *SD* = 3.58) as compared to infants before surgery (*M* = 5.06, *SD* = 3.83) in the upright condition (*t*(21) = -5.15, *p* < .001), but not in the inverted condition (*t*(21) = -1.86, *p* = .078, see Figure 5). As the P2 component reflects second order processing of the typicality of faces, such results reveal that infant faces were processed as more typical after repair surgery, suggesting that craniofacial repair surgery restores encoding of facial typicality that is more similar to that as a normal infant face.

Figure 6

Orientation x Palate Interaction for P2 Component

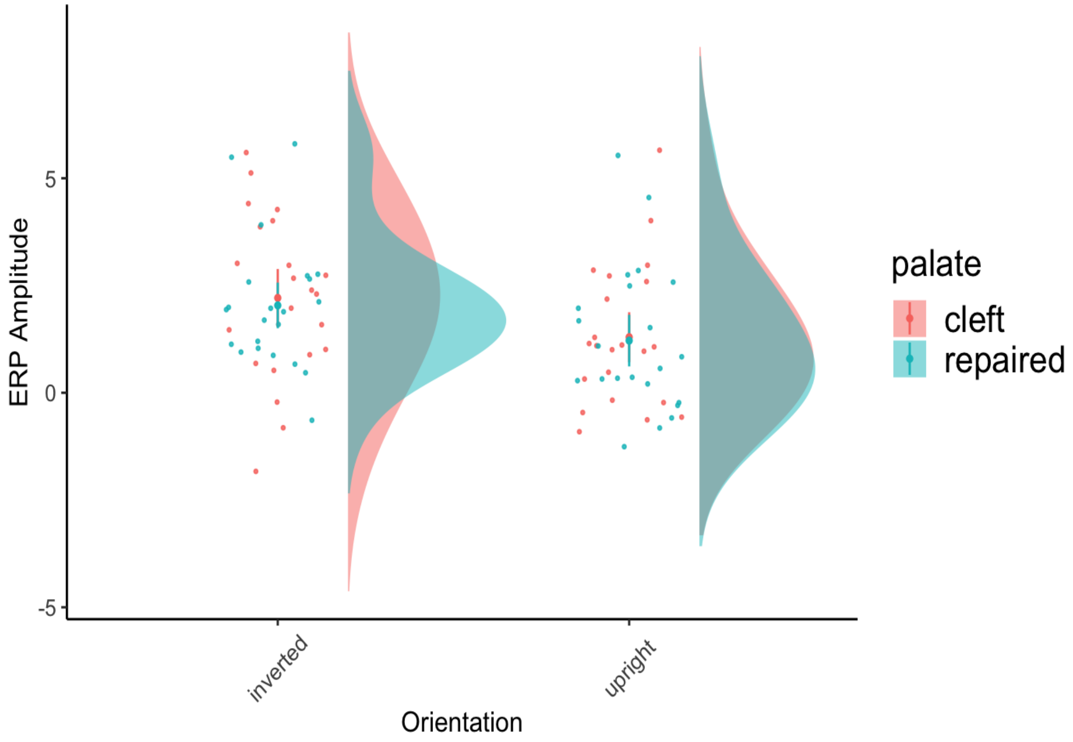


LPP

A main effect for orientation (*F*(1, 21) = 34.03, *p* < .001, η2 = .063) was observed, but not for palate (*F*(1, 21) = 0.94, *p* = .343, η2 = .001). There was no significant interaction between orientation x palate, *F*(1, 21) = 0.11, *p* = .738, η2 = .000. Such findings indicate that the emotional salience for infants affected by cleft lip/palate may remain the same even after craniofacial repair, suggesting that both the presence of the facial malformation and a repair scar evoke a similar emotional reaction from adults.

Figure 7

No Orientation x Palate Interaction for LPP Component



Discussion

The current study investigated whether craniofacial repair surgery restores more normative visual scanning and neural processing for faces of infants with cleft lip/palate. By using a cuteness rating task, as well as eye tracking and electroencephalography (ERPs), our findings replicate previous work suggesting that the presence of cleft lip/palate produces visual scanning and neural processing patterns that deviate from those observed for normal, unaffected infants. Furthermore, our findings also provide evidence suggesting that visual scanning and neural processing of infant faces who underwent repair surgery differ from their pre-surgery counterparts, and that the direction of these changes was more toward the visual scanning and neural processing seen for healthy, unaffected infant faces. This suggests that craniofacial repair surgery may restore more normative perceptual processing, although it remains unknown whether this surgical intervention *fully* restores "normal" processing. It is known that cleft lip/palate directly impacts caretaking behaviors, leading to less gaze behavior and reduced infant-caregiver bonding (Rayson et al., 2017; Sandberg et al., 2002). The current study’s findings support the notion that craniofacial repair surgery restores more normal responses to infants with cleft lip/palate, which suggests that craniofacial repair surgery may potentially reverse, or at least dampen, the adverse effects of cleft lip/palate on caretaking behaviors and parental investment may be fostered.

Findings from the current study suggest that, unsurprisingly, craniofacial repair surgery significantly increases perceived cuteness of infants with cleft lip/palate. Previous literature has indicated that the presence of cleft lip/palate negatively affects cuteness and has a direct impact on both visual and neural processing of faces (Hahn et al., 2023; Huffmeijer et al., 2018; Kringelbach et al., 2008; Parsons et al. 2011, 2013; Rayson et al., 2017). In addition, infants with cleft lip/palate typically are at risk for lower levels of nurturing behavior and less socially intimate engagement from caregivers (De Pascalis et al., 2017; Montirosso et al., 2012). The current study found evidence suggesting that the effects cleft lip/palate has on perceived cuteness are significantly improved by craniofacial repair surgery, which could also lead to significant improvements in caretaking responses. Indeed, based on Parsons et al.'s (2011) finding that the cuteness reduction due to cleft lip/palate reduced the motivational salience of infant faces, the current findings suggest that craniofacial repair surgery could potentially motivate greater caretaking behaviors given the previously established link between perceived cuteness and the reward value of infants (Hahn et al., 2015; Parsons et al., 2011; Yamamoto et al., 2009). Specifically, as repair surgery cosmetically fixes the physical appearance of the infant face, direct positive effects on parent-infant social bonding could be influenced, such as increased mutual gaze and more engagement during face-to-face interactions (Britton et al., 2001; Campbell et al., 2010; De Pascalis et al., 2017; Rayson et al., 2017; Sandberg et al., 2002). Our findings indicate that the change in eye gaze after craniofacial repair can, indeed, encourage greater perceptions of cuteness and potentially foster more joint attention and social expressiveness for affected infants (Britton et al., 2001; De Pascalis et al., 2017; Rayson et al., 2017). Cuteness has been described as a fundamental influence on human behavior, leading to increased attention, empathy, and prosocial behavior, and by surgically manipulating infant faces to reflect more of the kindchenschema attributes, potential benefits for motivated caretaking of affected infants can occur (Glocker et al., 2009; Kringelbach et al., 2016).

In line with previous findings suggesting that cleft lip/palate disrupts visual processing of infant faces (De Pascalis et al., 2017; Hahn et al., 2023; Rayson et al., 2017), the current study found that the mouth region of infant faces drew attention more rapidly and held attention longer for the pre-surgery as compared to the post-surgery faces. Considering the cognitive and social benefits of mutual gaze and direct, joint attention, the presence of a cleft lip/palate risks negative developmental outcomes for affected infants (Britton et al., 2001; Rayson et al., 2017). The current study's findings suggest that craniofacial repair surgery restores more normal visual scanning for infant faces with cleft lip/palate, which can in turn restore higher levels of caretaking behaviors. As heightened visual attention to the disfigured areas of faces is linked to negative emotional experiences and disrupted early infant-caregiver interactions (Murray et al., 2008; Stone & Potton, 2019), craniofacial repair surgery may be a way to reverse these effects (Sandberg et al., 2002). By altering attention capture and hold from the mouth region of the face to the eyes, less negative experiences and the promotion of intimate social and caregiving behaviors may be encouraged for infants who underwent craniofacial repair surgery.

Considering previous work (Hahn et al., 2023; Huffmeijer et al., 2018), the assumption that cleft lip/palate disrupts normative neural facial processing was supported by the findings of this study, as expected. Four ERP components were analyzed, including the P1, P2, N170, and LPP, and results mostly supported the expected hypotheses. The P1 component revealed no significant effects or interactions, as predicted. P2 amplitude, thought to reflect the later encoding of second order spatial relations of faces, in particular, facial typicality (Rossion et al., 2003; Schweinberger & Neumann, 2016), was found to be larger for infants after craniofacial repair surgery as compared to infants before surgery. Such results indicate that infants before surgery are perceived as further from prototypicality than faces after craniofacial repair, suggesting that craniofacial repair surgery allows for post-repair infants to be considered more typical during facial encoding.

The N170 was larger for infant faces before craniofacial repair surgery as compared to infants after surgery, in line with previous findings from Hahn et al. (2023). Although the direction of the effect is opposite from other research, such as Huffmeijer et al. (2018), the results demonstrate a disruption to normal configural processing. As a larger, more negative N170 response is typical for configural disruptions (Bentin et al., 1996; Carbon et al., 2005; Eimer, 2000; Itier & Taylor, 2004), our findings indicate the presence of cleft lip/palate is, indeed, a configural disruption. The lack of an interaction observed for the inversion of both infant faces before and after craniofacial repair surgery supports that it is equally as difficult to encode faces while inverted, regardless of the presence of a facial malformation. If an interaction had been observed, the disruption to neural processing would have been due to difficulties in featural processing, not configural, yet our findings indicate that this is not the case. These results suggest that reliance on additional recruitment of general object recognition processing mechanisms (Sadeh & Yovel, 2010; Yovel, 2016) is required while viewing infant faces with cleft lip/palate, but that after undergoing craniofacial repair, these neural processes are restored to a more normal level of, primarily, configural processing. Thus, it is indicated that the processing of infant faces before craniofacial repair uses different types of neural processing mechanisms, making it more difficult to process such faces. Craniofacial repair, however, at least partially reverses this effect by making it easier for adults to process infant faces who underwent repair surgery. Furthermore, as there is evidence that adverse effects on configural processing of infants with cleft lip/palate can be reversed on a neural level, it can be assumed that the negative behavioral effects from caregivers may be reversed as well.

The LPP component showed no differences between infants before and after craniofacial repair surgery. These findings do not support the current study's hypothesis, or the previous findings from Hahn et al. (2023), yet they suggest that the evidence of surgical intervention on the face of infants may evoke intense emotional reactions. However, it is not possible to know whether the lack of difference is due to a low level of emotional activity for both faces, or a high level of activity for both faces. Future studies may take advantage of adding a control group to further investigate these subtle effects. Despite this, it is assumed that because there is still a slight indication of a facial malformation, emotional reactivity is amplified the same for infants who underwent craniofacial repair as compared to infants before surgery. Considering the precise interventions which make up infant lip or palate repair, including the creation of an intact upper lip with appropriate facial aesthetic, the repair of underlying muscular structures, the repair of hard and/or soft palates, and potential treatment of associated nasal deformities (Campbell et al., 2010), an elevated emotional reaction to a cleft lip/palate repair scar can be explained. Additionally, as the LPP has been associated with a commitment of motivational and attentional circuits in the brain to affective visual stimuli, it is also context dependent (Hajcak et al., 2009; Liu et al., 2012; Schupp et al., 2000). Thus, the valence of the emotion being experienced during periods of increased LPP response cannot be directly identified, although the levels of emotional arousal are represented (Schupp et al., 2000). Overall, findings demonstrate that the disruption of normal facial configuration by cleft lip/palate significantly impacts early visual and neural responses, and that craniofacial repair surgery has the potential to restore more normative processing for affected infants.

While there are many potential benefits for this research, there are also several limitations to consider when drawing conclusions about whether craniofacial repair surgery restores normative facial processing for infants with cleft lip/palate. First, it is critical to note that stimuli only included photographs of infants before and after craniofacial repair. Thus, there was no healthy and/or normal infant face with the same identity to compare with. To better explore whether repair surgery completely restores normal processing, future research should analyze these same visual and neural patterns, but with a stimulus set of infants with normal, cleft lip/palate, and repaired faces with the same identity. With this, the visual and neural patterns observed for post-surgery infants can be compared directly to the patterns observed for their same faces but with a normal appearance. By making this comparison, a stronger argument can be claimed regarding the restoring capabilities of craniofacial repair surgery. Second, it is also important to recognize that post-surgery faces are always older than pre-surgery faces. Considering the age effect, it may be possible that adults are rating infants either less or more cute relative to their age, as it affects the ability to recognize and distinguish between faces. Future studies should determine if and how age may influence responses to these infant faces.

Findings suggest that craniofacial repair surgery has been found to restore more normal visual and neural processing for infants with cleft lip/palate. This occurs as craniofacial repair surgery has been shown to significantly increase perceived cuteness of affected infants by restoring characteristics closer to those of the kindchenschema phenomena. As cuteness is an innate releasing mechanism for caretaking behaviors (Glocker et al., 2009; Kringelbach et al., 2006), it can be assumed that the promotion of caregiving behaviors will occur post repair surgery as these infants are rated as much cuter as compared to before surgery. Craniofacial repair surgery also restores more normative visual scanning of infant faces, such that visual attention to the mouth is less capturing for infant faces after surgery as compared to before surgery. With the understanding that mutual gaze and joint attention at the eyes facilitates infant-caregiver interactions and social bonding (Britton et al., 2001; Rayson et al., 2017), it can be expected that the negative effects of the facial malformation may be reversed with craniofacial repair surgery. For example, Murray and colleagues (2008) found that affected infants who undergo repair at later stages are at risk for impaired cognitive developments due to difficulties in mother-infant interactions. Our data suggests that craniofacial repair alters the basic face processing strategies that may contribute to such difficulties due to the lack of eye contact and impaired configural processing mechanisms, and therefore, may promote more intimate infant-caregiver interactions.

Finally, craniofacial repair significantly impacts both the early (N170) and late (P2, LPP) neural processing of infant faces. With established easier configural processing and a more “face typical” appearance, less effort to process infant faces who underwent craniofacial repair can be expected. This restorative function of craniofacial repair on the underlying neural mechanisms of facial processing may in turn encourage stronger parental and caregiver bonds and engagement with infants who received the surgery. Essentially, it can be assumed that with the help of craniofacial repair, adverse cognitive, behavioral, and social effects can be reversed, and more normal visual and neural processing of these infant faces takes place. This study provides a neurobiological explanation as to why early infant-caregiver interactions for infants with cleft lip/palate can be disrupted, and more importantly, is a first step in determining the effectiveness of surgical interventions on the neural mechanisms of infant face processing.

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