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Report of Altered Sensation in Patients With Cleft Lip

Dr. Greg K. Essick, D.D.S., Ph.D. [Professor],

Department of Prosthodontics and Curriculum in Neurobiology, University of North Carolina, Chapel Hill, North Carolina

Mr. Craig Dorion, B.S. [Research Assistant],

Department of Orthodontics, University of North Carolina, Chapel Hill, North Carolina

Mr. Seth Rumley, B.S. [Research Assistant],

University of North Carolina, Chapel Hill, North Carolina

Ms. Lyna Rogers, R.D.H., B.S. [Research Analyst],

Department of Orthodontics, University of North Carolina, Chapel Hill, North Carolina

Mr. Michael Young, B.S. [Research Technician], and

University of North Carolina, Chapel Hill, North Carolina

Dr. Carroll-Ann Trotman, B.D.S., M.A., M.S. [Associate Professor]

Department of Orthodontics, University of North Carolina, Chapel Hill, North Carolina

Abstract

Objective—To determine whether patients with cleft lip have normal perioral sensation.

Design—Each subject was carefully questioned about the following: sensation in the face at rest, light touch of different areas, and sensation in natural situations (e.g., exposure to cold weather) that reveal sensory abnormalities. A cotton-tip applicator stick was stroked lightly across the facial skin. The subject's descriptions of the evoked sensations were used to identify and outline areas with abnormal sensation.

Setting—Data were obtained from subjects participating in a longitudinal, university-based study of the functional outcomes of lip revision surgery.

Patients—Seventeen patients with cleft lip and 12 control subjects (aged 7 to 22 years, mean 12.9 years) participated.

Results—In contrast to control subjects, 9 of 16 patients (56%) reported loss in sensation, described as decreased touch, scratch, tickle, or tingle intensity. Six other patients (38%) reported additional sensation, described as increased scratch, tickle, or tingle intensity. In eight patients, the altered sensation was restricted to the skin area flanking and including the visible scar, encompassing no more than 25% of the total area bound by the inferior nose, nasolabial grooves and inferior vermilion. In seven patients (unilateral cleft), the altered area extended to the contralateral, noncleft side of the upper lip or onto the philtrum.

Conclusions—In contrast to the literature, sensation in the upper lip of many patients with cleft lip is not normal. Loss in sensation is exhibited most commonly and limited largely to the skin overlying tissues traumatized during reconstructive surgery.

Keywords

cleft lip; craniofacial; impairment; sensation; sensory; testing

The studies published to date suggest that patients with cleft lip and palate exhibit normal or near normal orofacial sensory function (Posnick et al., 1994a, 1994b; Uchiyama et al., 1998; Akal et al., 2000). Tactile sensitivity (pressure detection thresholds) and acuity (two-point discrimination) are comparable for patients and control subjects on the upper lip, the midface, and the hard and soft palates (Posnick et al., 1994b; Uchiyama et al., 1998; Akal et al., 2000). Similarly, 1 year following reconstructive surgery, sensitivities to pin prick, warmth, and cold return to normal on the upper lip and nose, both on native skin and transplanted skin from pedicle graphs (Takahashi and Kato, 1966; Akal et al., 2000). Although subtle alterations in vibrotactile sensitivity (amplitude detection thresholds) have been reported, their interpretation is unclear. For example, decreased sensitivity to vibration has been observed on the hard palate at the premolar teeth, suggesting injury to the greater palatine nerve (Uchiyama et al., 1998). In contrast, increased sensitivity has been observed on the nasolabial and labial skin (Posnick et al., 1994b). Perhaps the most clinically important evidence of normal sensory function in these patients is the paucity of reports from adolescent and young adult patients of altered sensation on the face or in the mouth (Takahashi and Kato, 1966; Posnick et al., 1994a, 1994b; Akal et al., 2000).

These findings are encouraging from a clinical standpoint and could be viewed as reason to abandon research on sensory function in patients with cleft lip (Posnick and Grossman, 2000). However, based on the incidence and severity of sensory impairment in patients after orthognathic surgery (Essick et al., 2001, 2002) or facial trauma (Stranc and Fogel, 1984; Stranc et al., 1987), the lack of altered sensation in patients with cleft lip is unexpected and thus merits continued investigation for this reason. To this end, an exploratory pilot study was conducted to probe the subjective nature of sensation in young patients with cleft lip. Rather than administering conventional neurosensory tests at fixed skin sites, the examiner talked with the patient about “how the face feels,” both when touched and in natural situations such as exposure to cold weather. Cotton-tip and wooden-stick stimuli were stroked across the face. The subject’s descriptions of the evoked sensations were used to locate and outline areas on the face that did not have normal sensation. With use of these procedures, evidence of altered sensory function on the upper lip was identified in many patients but not in control children without cleft.

Methods

Patients

The data reported below were obtained from subjects who were participating in a longitudinal study of the functional outcomes of lip-revision surgery. The ongoing study includes assessment of facial mobility, perioral fine motor control, and facial muscle and sensory function. Seventeen patients (10 boys and 7 girls, aged 7.1 to 21.7 years, mean 13.2, SD 3.9) and 12 noncleft control subjects (six boys and six girls, aged 7.5 to 20.8 years, mean 12.5, SD 3.9) were studied in preparation for this report. Nine patients (53%) had unilateral cleft lip and palate, four patients (24%) had bilateral cleft lip and palate, and four patients (24%) had unilateral cleft lip only. No subject had undergone surgery to the lips, face, or mouth within 24 months of testing. Data were obtained from each subject during a half-day-long appointment. The time for this subjective sensory evaluation varied from about 10 to 40 minutes, depending on the cognitive abilities and cooperation of the patient. The study was approved by the Committee on Investigations Involving Human Subjects at the University of North Carolina School of Dentistry.

Interview

The subject was comfortably seated in the clinical laboratory. Three sets of questions pertaining to alterations in sensation and the functional consequences were asked. The exact wording of the questions and the time allowed for responses varied according to the examiner's judgment of the patient's cognitive abilities, cooperation, and attentiveness. The interview process was refined through experience gained from administration of the questions to different subjects. As such, not all questions described below were asked of patients who were tested early in the study. Moreover, select questions were reworded and asked later during the interview to confirm consistency of response and the attentiveness of the subject. A summary of the subject's responses was written at the end of testing.

The first set of questions sought to identify evidence for mechanical hypoesthesias, hyperesthesias, and paresthesias. The subject was instructed to lightly stroke his fingers over the face and lips and while doing so to indicate any area that "felt different." Examples of "feels different" were provided and included the following: lighter in touch or pressure, greater in touch or pressure, loss of feeling, numbness, pins-and-needles-like feelings, tingling, and tickling. For those subjects who had received dental anesthesia in the recent past, "numb" and "pins-and-needles-like" were further described as feelings similar to those experienced during the anesthesia. Building on the possibility of paresthesias, the patient was asked whether he could contract the facial muscles to make pins-and-needles-like feelings or tingling feelings in the face, the upper lip, or the lower lip.

The second set of questions sought to identify alterations in temperature sensitivity and touch intensity that might be detected during natural stimulation. The subject was asked whether exposure of the face to cold weather, cold water (such as in shower or swimming pool), or cold foods (such as ice cream) made any part of his face or lips feel different from other parts. Similar questions were posed for exposure of the face and lips to hot weather, hot water, and hot foods (such as hot soup). Differences reported for foods were investigated further to determine whether the sensations experienced were commensurate with cold and hot substances touching the lips or were abnormal in intensity, distribution, or persistence, suggesting impairment. Finally, the subject was asked whether air blowing on the face (wind or fan) made any part of the face or lips feel different from other parts. Exposure to outdoor wind while playing was addressed to evaluate combined light tactile and thermal stimulation of the facial tissues.

The third set of questions sought to identify functional difficulties that might relate to alterations in sensory function. The subject was asked whether there was anything that he could not do as well with his lips and face as other people (e.g., his friends at school). He was further asked whether any area on the face affected his speech, singing, whistling, or smiling. The examiner asked additional questions to explore all positive responses.

Mapping of Area of Altered Sensation

After the interview, areas of altered sensation were identified by stroking stimulus objects lightly across the face, mid-cheek to midcheek along parallel paths from the inferior nose to the mentolabial groove. The stimulus objects consisted of the cotton tip and the wooden ends of a cotton-tip applicator stick. The subject was asked to concentrate and tell the examiner of any change in how the face or lips felt. The examples of different types of feelings, as previously described, were provided. In addition, it was noted that the stimulus might feel smoother, rougher, duller, sharper, scratchier, or less scratchy. Highly motivated subjects were given the opportunity to stroke and use the stimuli to indicate suspect areas. A mirror was provided to assist in this process. The examiner outlined the identified areas with

a washable-ink marker and photographed the subject's face with a digital camera. The outlines were accentuated for publication purposes (Figs. 1 through 4).

Analysis

The frequencies of positive responses to the same questions were tabulated for the patients and the subjects without cleft. In the absence of sensory impairment, no positive responses were expected. Positive responses from six or more patients with cleft lip suggested a statistically significant group difference (Fisher's exact test, $p = .05$, two tailed).

Results

General Observations

The subjects were generally very receptive to the study procedures. The most detailed and complete responses were provided by subjects 10 years of age and older. The older subjects were also particularly helpful during the mapping exercise and notified the examiner if the boundaries had not been drawn to the appropriate locations on their face. In the authors' opinion, meaningful data were obtained from all subjects with the exception of two of the youngest subjects. The youngest patient with cleft lip (age 7.1 years) was easily distracted and totally inattentive to the questions. He responded "no," if at all, to the questions and was not receptive to talking about his face.

In contrast, one of the youngest subjects without cleft (age 8.6 years) was very talkative and provided positive, but inconsistent, responses to most questions. For example, he reported that ice cream was colder on his upper lip, soup was hotter on his upper lip, but shower water was hotter on his lower lip. He also reported that wind felt different on his cheeks, making them feel smoother than the rest of his face. However, the cotton stimulus felt smoother on his upper lip precisely over the anatomical region bound by the two nasolabial grooves. The meaningless data from these two subjects were not included in the results presented below.

Sensory Alterations in Subjects Without Cleft

The subjects without cleft were recruited and studied after the full battery of questions for the interview had been developed. Their responses did not suggest any sensory or motor impairment. No subject reported that any one area of the face felt different from another area on touching his skin. No alteration in sensation was detected or could be mapped by stroking the skin with the stimuli. Cold, heat, or air did not make any area of the face feel different from any other area. Two subjects remarked that cold weather made both their upper and lower lips feel numb. No functional deficiencies were noted. Two subjects reported difficulty in enunciating "r" sounds.

Sensory Alterations in Patients With Cleft Lip

At the beginning of the interview, 3 of 16 patients (girls, aged 11.6, 16.1, and 21.6 years) reported decreased sensation in the upper lip on touching and examining their face. The remaining 13 patients (81%) indicated that no area on the face felt different from any other area. On additional questioning, 3 of these 13 patients (aged 8.1, 10.5, and 16.5 years) noted that their upper lip was more sensitive to cold, heat, or both, and one male patient (aged 10 years) noted that wind made his upper lip tickle. All considered, 7 of 16 (44%) of the patients provided responses consistent with altered sensation limited to the upper lip.

Compared with the interview, stroking the face with the stimuli was more effective and identified areas of altered sensation in 15 of the 16 patients (94%). The 15 patients included all seven patients who reported an alteration during the interview. Use of the cotton stimulus

identified altered sensation in 13 of these patients (87%). The findings were a decreased touch or tickle intensity (eight patients) or an increased tickle or tingle intensity (five patients). Use of the wooden stimulus identified altered sensation in 10 of the 15 patients (67%). These findings were a decreased touch or scratch intensity (six patients) or an increased touch, scratch, or tingle intensity (four patients). Of the 10 patients tested with both cotton and wooden stimuli, similar alterations (e.g., decreased touch and scratch sensation, respectively) were revealed in seven patients. For two subjects, altered sensation was evoked by only the cotton stimulus or the wooden stimulus. Although no subject described the sensation as “pins and needles-like,” five patients chose the word “tickle” and two chose the word “tingle” to describe the alteration. All totaled, 9 of the 16 patients (56%) exhibited loss in sensation (decreased touch, scratchiness, tickle, or tingle) as determined from responses to the stimuli stroked across the skin. Six of the 16 patients (38%) exhibited an increase in sensation (increased tickle, tingle, or scratchiness).

With only one exception, the 15 areas of altered sensation were limited to the upper lip/inferior nose and included the visibly scarred tissue in whole or part. For nine patients, the area included scarred tissue on both hairy skin and vermillion; for three patients, on hairy skin only; and for three patients, on vermillion only. In eight patients (53%), the altered area was restricted to the skin flanking and included the visible scar, extending no farther than about 0.5 cm to either side of the scar. As a rough estimate, the area encompassed no more than 25% of the total area bound by the inferior nose, nasolabial grooves, and inferior vermillion. These areas of altered sensation in two of the eight patients are illustrated in Figure 1. Figure 1A shows the mapped area for a 16-year-old boy with bilateral cleft lip and palate. The patient reported that he did not feel the cotton stimulus “as much” inside the bound area as outside or on the lower lip. The wooden stimulus was described as “much less scratchy” and proved more effective than the cotton stimulus in distinguishing impaired from normal sensation. Figure 1B shows the mapped area for a 16-year-old girl with a right unilateral cleft lip. The patient reported that the cotton stimulus felt more ticklish in the area extending from her nostril to the dimple in her vermillion. Similarly, the wooden stimulus caused a greater scratchy sensation in the bound area than on the contralateral, noncleft side.

In seven patients (47%), all with unilateral clefts, the altered area extended beyond the visible scar onto the philtrum and for five patients farther onto the contralateral side of the upper lip. The area in these patients most often encompassed 25% to 50% of the area bound by the inferior nose, nasolabial grooves, and inferior vermillion. Such areas of altered sensation in two of the seven patients are illustrated in Figure 2. Figure 2A shows the mapped area for a 21-year-old woman with a right unilateral cleft lip and palate. The patient reported a uniform, distinct loss of sensation while touching her skin. On mapping with the stimuli, she summarized the sensations evoked in the bound area as “less, less scratchy, less tingling, numb.” She was the only patient to report a (partial) loss of the sense of warmth and cold in the upper lip. Figure 2B shows the mapped area for a 10-year-old girl with a left unilateral cleft lip. In response to the stimuli, the patient reported that she “could not feel as much” inside as outside the bound area or as on the lower lip. No area was anesthetic, however, and pressure applied directly over the visible scar caused a greater sensation than pressure to the adjacent skin.

Of the 12 patients questioned about function, nine stated that the cleft had no impact on their speech, facial expression, or other behaviors. Two indicated cleft-related difficulties in speech, which were apparent to the examiners. One of these subjects exhibited greater sensory alteration than observed in most subjects in this study (mapped area shown in Fig. 2A). A third subject (aged 16.5 years) thought that his face did not feel normal during smiling and had difficulty whistling (mapped area of sensory alteration shown in Fig. 1A). Another patient (aged 10.1 years) could not detect food remaining on the repaired tissue,

according to her mother. Three of the 16 patients reported that cold weather made their upper lip numb and difficult to move.

Variation in Repeated Mappings

Before and during the course of the longitudinal study in which the subjects participated, the mappings were repeated for a few patients. Comparison of the outcomes made evident the need for a careful and systematic approach in mapping the face. For example, Figure 3 shows two mappings by the same examiner of a 16-year-old girl with a left unilateral cleft lip and palate. The mapping at top (Fig. 3A) was obtained during an examination prior to initiation of this study. By lightly prodding the skin with a sharp dental explorer, areas of altered sensitivity were identified and outlined (Essick, 1992). On the area to the patient's left, pinprick was consistently described as "dull," compared with "sharp" on the lower lip. On the area to the patient's right, pinprick felt abnormal, but the sensations were inconsistent: On some proings it was dull; whereas on others, it was sharper than on the lower lip. Two weeks later, the mapping was repeated more carefully with the cotton-tip applicator stick as part of this study (Fig. 3B). A similar but slightly larger area of altered sensation with irregular borders was identified. Distinct zones of hyposensitivity and hypersensitivity were located, explaining the variable responses to the dental explorer when probed previously at random locations.

Shown in Figure 4 is a second example, two mappings of a 16-year-old boy with a left unilateral cleft lip and palate. The first mapping was performed by examiner G.E. and revealed an increased ticklish sensation to the cotton stimulus in the vicinity of the cleft scar (Fig. 4A). The ticklish sensation was consistently evoked on the vermilion area outlined in the photograph. The second mapping, performed by examiner C.D. 3.5 months later, also revealed an increased ticklish sensation to the cotton stimulus but consistently so on the hairy skin surrounding the scar (Fig. 4B). Examiner C.D. was blinded to the mapping of examiner G.E. Differences in the areas from which consistent responses were obtained likely were due to subtle differences in the pressures or manners in which the examiners applied stimuli to the skin. The cotton stimulus did not evoke an abnormal ticklish sensation on other areas of the upper lip, lower lip, or face during the mappings. The wooden stimulus did not evoke altered sensations at any site during either examination.

Discussion

With only one exception, the patients with cleft lip and palate in the present study did not have normal sensation on the upper lip. For many patients the area of altered sensation was mild and restricted to the tissues traumatized during primary and secondary revision surgeries. However, for about half of the patients, it extended to the contralateral, noncleft side of the upper lip or onto the philtrum. In adults, injury and reconstructive surgery of the lips often result in a loss of sensation, which is frequently accompanied by impaired labial motor function (Stranc and Fogel, 1984; Stranc et al., 1987). Similar neurosensory sequelae of reconstructive surgery of the lips in children seem plausible, barring the possibility of a more complete or perfect regeneration of injured nerves in younger patients. Additionally, scar-associated alterations in tissue compliance might affect how, and which, skin receptors respond to tactile stimuli. Because of the loss of innervation, changes in tissue compliance, or both, one would predict altered sensation in cleft scars, although this has not been reported in the literature or studied previously to the authors' knowledge. In the present study, the presence of scarring might explain why some patients described the wooden stimulus as "more scratchy" over the repaired tissues and why one patient reported that his face did not feel normal during smiling.

The findings of this study are in contrast to those of previous studies that reported no alterations in sensory function (Posnick et al., 1994a, 1994b; Uchiyama et al., 1998; Akal et al., 2000). Three possibilities might explain the failure to detect impairment in lip sensation. First, sensory function was evaluated at fixed skin sites only in the previous studies. Given that individual patients exhibit uniquely different repaired and scarred tissues, the specific sites tested might not have included the patients' areas of altered sensory function.

A second plausible reason that previous studies found no sensory alterations is that only objective neurosensory testing procedures were employed (Essick, 1992). During these conventional procedures, patients are asked to respond when, or if, they feel contact with the skin, vibration, two points of contact, warmth, pain, etc. (Essick, 1992; Posnick et al., 1994b; Uchiyama et al., 1998). Their responses are accepted as accurate reflections of their sensations. These methods are preferred for measuring the absolute and differential sensitivities of sensory processing, but they are largely insensitive to how stimuli feel qualitatively on the skin (Essick, 1992; Essick et al., 1999). With the return of normal touch, temperature, and pain sensibilities after reconstructive lip surgery, stimuli do not always feel the same on affected skin as on the adjacent normal skin (Takahashi and Kato, 1966). This additional insight can be obtained only by asking the patients to compare and describe the sensations evoked by the same stimuli on suspect and unaffected skin sites.

It would be informative to compare the subjective evaluations afforded by the procedures described in this article with the outcome of conventional neurosensory testing at fixed skin sites. It is possible that the patients in this study would appear to have normal sensory function based on objective testing. This issue will be addressed at the end of the longitudinal study at which time two-point discrimination and thermal threshold data will be available.

A third factor limiting the identification of sensory alteration in patients with cleft lip is their altered perception of normal sensation. That is, unless questioned carefully, patients might not recognize as abnormal those sensations they have had since infancy, particularly as sensation in the upper lip is likely to be secondary to esthetic and functional concerns. That this is the case was suggested by the findings of this study. At the beginning of the testing session, only 3 of the 16 patients responded affirmatively when questioned as to alterations in sensation. However, 15 of the 16 patients noted altered sensation in response to the stroking movements of the applicator stick during the mapping exercise. It is noteworthy that two of the three patients who initially reported alterations in sensation were girls aged 16 and 21 years. It might be that these patients had acquired better introspection skills and awareness of facial sensation from the application of facial cosmetics.

The altered sensations (both losses in sensation and hypersensitivities) reflect abnormalities in sensory information from the upper lip or in the central neural processing of that information (Essick, 1992; Essick et al., 2002). Sensory information from the face serves a role in proprioception, as well as in perception, and affects, for example, the manner in which speech movements are achieved (Putnam and Ringel, 1972; Edin and Johansson, 1995; Trulsson and Essick, 1997; Tremblay et al., 2003). Altered patterns of sensory information because of loss of receptors or changes in skin compliance are predicted to have a negative impact on the motor control of the face, even in cases for which speech appears normal. Such abnormalities in children with cleft lip have been demonstrated and include altered electromyographic responses evoked in the facial musculature by mechanical taps applied to the lips (D'Antonio et al., 1994, 1995). To what extent these abnormalities contribute to clinically observable dysfunction in facial movements is unclear (Trotman et al., 2000).

The relationship between altered sensation and clinically observable dysfunction is likely very complex. Of the two subjects who exhibited the most pronounced sensory alteration, one exhibited severe speech impairment (mapped area shown in Fig. 2A), and one exhibited near normal speech (mapped area shown in Fig. 3). Interestingly, the latter subject reported that she had worked hard to learn to speak clearly. Sensory retraining, both through professional therapy and self-teaching, is a means by which young patients can learn to utilize altered patterns of sensory inputs from their lips and in doing so, minimize the presentation of functional impairment. In this regard, no disabling motor deficits were identified in the present study that could be attributed largely to loss in sensory function (Stranc and Fogel, 1984; Essick, 1998; Posnick and Grossman, 2000). One such deficit, drooling, is often observed in adults after orthognathic and lip surgeries. The adults may have greater sensory impairment than young patients with cleft lip or have less capacity for sensory retraining.

Collection of the data in the present study required the cooperation, concentration, and active participation of young subjects in a clinical environment. As such, the data were necessarily biased by the cognitive skills of the individual patients and their selection and use of words to describe the impairments. The mapping data were also biased by the precise manner in which the two examiners applied tactile stimuli to the face. The consistency across subjects, however, makes evident that the major finding of this study, that altered sensation often exists in the cleft lip, cannot be attributed to these biases.

In conclusion, many patients with cleft lip and palate have areas of altered sensation on the upper lip. This is observed clinically on careful questioning of patients about the “feelings” in their lips and face. Within the scarred areas, the altered sensation can be evoked by light, brushing contact with the fingertip or the ends of a cotton-tip applicator stick. Although loss in sensation is more likely, some patients may report increased sensations of scratchiness, tingle, or tickle.

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FIGURE 1.

Areas of altered sensation in two of the eight patients for whom the mapped area was limited to the skin flanking and including the visible scar. A: A 16-year-old boy with bilateral cleft lip and palate. B: A 16-year-old girl with right unilateral cleft lip.

FIGURE 2.

Areas of altered sensation in two of the seven patients with unilateral cleft for whom the mapped area extended contralaterally onto the noncleft side or the philtrum. The areas also extended onto the nose. A: A 21-year-old woman with right unilateral cleft lip and palate. B: A 10-year-old girl with a left unilateral cleft lip.

FIGURE 3. Repeated mapping by the same examiner of a 16-year-old female subject with a left unilateral cleft lip and palate. A: Quickly performed mapping with use of a dental explorer. B: More carefully performed mapping 2 weeks later with cotton-tip and wooden-stick stimuli.

FIGURE 4.

Repeated mapping of a 16-year-old male subject with a left unilateral cleft lip and palate. A: First mapping by examiner G.E. B: Second mapping 3.5 months later by examiner C.D. See text for additional details.