

A novel method for evaluating postsurgical results of unilateral cleft lip and palate with the use of Hausdorff distance: presurgical orthopedic treatment improves nasal symmetry after primary cheiloplasty.

ハウスドルフ距離を用いた片側唇裂手術後の鼻孔形態の新しい評価方法：術前顎発育補助装置の使用の有無と初回唇裂手術後の鼻の形態改善に関する分析

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1. Introduction

When performing primary cheiloplasty for cleft lip and palate, the reconstruction of attractive nose and lips is an important concern of the plastic surgeon. In addition, since the shape of the nose in unilateral cleft lip and palate is the biggest concern for the family ¹, how to adjust the shape of the nose at the primary cheiloplasty is another critical issue. In recent years, equipment such as presurgical orthopedics have improved the reconstruction of attractive features ²⁻⁴. However, the surgical outcome with respect to the shape of the nose is difficult to evaluate, because of the complexity of its shape.

Various methods for evaluating facial appearance have been reported. These include measurements based on the anthropometric point ⁵⁻⁷, measurement of the area or aspect ratio of the naris ⁸, and acquisition of the facial appearance as 3-dimensional image data ⁹⁻¹¹. The shape created by the surgery has a complex structure, and even if the area, aspect ratio, or angles are the same on the affected and unaffected sides, the similarity of their appearance is not always compatible with judgment made by the human eye. Therefore, various analyses adding more reference points or measurements to assess the morphology in detail have been reported ^{7, 12-15}. However, there is no report comparing the affected to the unaffected side post-operatively by direct digitalization.

Here we propose a new method for evaluating the similarity of the nasal cavities

between the unaffected and operated side for unilateral cleft lip and palate patients after cheiloplasty, by quantifying the geometry of the nasal cavity. For this analysis, we focused on the symmetry of the nasal cavities as a final goal of the operation, and measured the Hausdorff distance to assess this outcome quantitatively.

The Hausdorff distance is defined as follows;

Let X and Y be two non-empty subsets of a metric space (M, d) . We define their

Hausdorff distance $d_H(X, Y)$ by

$$d_H(X, Y) = \max \left\{ \sup_{x \in X} \inf_{y \in Y} d(x, y), \sup_{y \in Y} \inf_{x \in X} d(x, y) \right\}$$

where *sup* represents the supremum and *inf* the infimum.

Equivalently

$$d_H(X, Y) = \inf \left\{ \epsilon > 0; X \subseteq Y_\epsilon \text{ and } Y \subseteq X_\epsilon \right\}$$

where

$$X_\epsilon := \bigcup_{x \in X} \{z \in M; d(z, x) \leq \epsilon\}$$

that is, the set of all points within ϵ of the set X (sometimes called the epsilon-neighborhood of X).

The Hausdorff distance is a virtual distance that measures how far two subsets of a metric space are from each other; it is not the actual distance that is measurable in SI

units. If two objects are identical, the score is 0, and the score becomes larger as the shapes of the two objects become more different. The Hausdorff distance is used as a computer algorithm for recognizing patterns ¹⁶, and in medicine, it is used for recognizing structures on CT ^{17,18} or MRI ¹⁹ images, and for analyzing the morphology of developing neurons ²⁰, blood vessels of the fingers ²¹, and other structures ²².

In this study, we propose a new method for evaluating the shape of the nares after cheiloplasty by measuring the Hausdorff distance. We also used this method to evaluate the effectiveness of using a Hotz plate for retaining the shape of the naris after primary cheiloplasty.

2. MATERIALS AND METHODS

2-1. Patients

We studied 30 patients with unilateral cleft lip and palate who received unilateral cheiloplasty at the Tsukuba University Hospital between 2000 and 2011. All the patients were Japanese infants; 10 were male and 20 were female. None of the cases had complications other than this disorder. Of these 30 cases, 15 were treated without using a Hotz plate (nHP-group) and 15 were treated using a Hotz plate, between 2005 and 2008 (HP-group). The Hotz plate was used for patients whose parents wanted it after 2005. All the patients underwent cheiloplasty at 3 months by a modified rotation-advancement method with anatomical reconstruction of the orbicular oris muscle and with nasal cartilage correction. The same surgeon performed all the surgeries. A series of frontal view photographs was taken for each patient one month after the primary cheiloplasty. All the photographs were taken using a standardized handheld technique by the same investigator with the same digital camera.

2-2. Surgical technique

The surgical procedure was a modified rotation advancement cheiloplasty with nasal cartilage correction in all infants. The alar base was dissected from the underlying bony

structure in the plane above the periosteum. The orbicularis oris muscle was reconstructed by basket-weave muscle repair²³. After all the muscles and flaps were sutured, the nasal cartilage was corrected. Minimum subcutaneous undermining was carried out over both alar cartilages by reverse-U incision²⁴. The alar cartilage of the cleft side was then sutured to the alar cartilage of the non-cleft side and to the lateral cartilage.

Based on the post-operation photograph, we standardized the object and performed the following measurements and analyses.

2-3. Measurement of Hausdorff distance

- i) The outline of the naris was plotted using Canvas 11J with GIS (ACD Systems International Inc. Seattle, USA), and the shape of the naris on both sides was extracted (Fig. 1A).
- ii) The internal canthal distance was fixed to 100 mm on the graphic software as a horizontal baseline, and all cases were standardized to it.
- iii) The outline of the affected side was then flipped horizontally (Fig. 1B).
- iv) The data were saved as an SHP file to be converted to the geographic information system (GIS).

v) The SHP files were then incorporated into PostGIS 1.5.1, an open-source software program that adds support for geographic objects to the PostgreSQL 8.4.8 object-relational database. In parallel, the center-of-mass coordinate for each case was assigned as (0,0) using a function called “ST_Centroid”, and the Hausdorff distance of the shape of both nares was measured using a function called “ST_Hausdorff Distance” in PostGIS.

(Note that PostGIS "spatially enables" the PostgreSQL server, allowing it to be used as a backend spatial database for GIS.)

2-4. Visual evaluation

The shape of the nares extracted in 2-3.-i) in all 30 cases was classified into 5 categories based on their symmetry, determined by the visual evaluation of 6 experts in plastic and reconstructive surgery and oral and maxillofacial surgery. The criterion was the symmetry of the size and shape of the left and right naris. Each evaluator assigned 6 cases to each category. The score was determined as the average of all the evaluators' scores.

2-5. Area ratio, perimeter ratio, aspect a/u ratio

Based on the shape of the naris extracted in 2-3.-i), we measured the area and perimeter of the naris with Image J (NIH, Bethesda), then calculated the ratio between the affected and unaffected sides to determine the area ratio and perimeter ratio, respectively. For the aspect ratio, we used Feret's diameter to measure the maximum axis as the long axis and minimum axis as the short axis, and calculated the (long axis)/(short axis) ratio. The aspect a/u ratio was determined as the aspect ratios of the affected / the aspect ratios of the unaffected sides.

2-6. Statistical analysis

The correlation coefficients among the area ratio, perimeter ratio, aspect a/u ratio, visual evaluation, and Hausdorff distance data were analyzed by Pearson's correlation coefficient test. The scores of the area ratio, perimeter ratio, aspect a/u ratio, visual evaluation, and Hausdorff between the HP and nHP groups were analyzed by Student's t-test. Statview 5.0 (Abacus Corporation, Baltimore) was used for the statistical analyses. A P-value < 0.05 was considered significant.

3. RESULTS

3-1. Correlation among different evaluations

Table 1 shows the results of the visual evaluation, Hausdorff distance, area ratio, perimeter ratio, and aspect a/u ratio for all the patients. The coefficient of correlation between each method is shown in Table 2. Significant correlation with the visual evaluation was observed for the Hausdorff distance (correlation coefficient: $r = -0.805$, $P < 0.001$) and aspect a/u ratio (correlation coefficient: $r = -0.470$, $P < 0.01$). Significant correlation was also observed between the area ratio and perimeter ratio (correlation coefficient: $r = 0.642$, $P < 0.001$) and between the perimeter ratio and aspect a/u ratio (correlation coefficient: $r = 0.463$, $P < 0.01$). There was no correlation between other pairs of methods. Regarding the correlation between the area ratio and perimeter ratio, the perimeter will increase as the area becomes larger. Regarding the correlation between the perimeter ratio and aspect a/u ratio, the perimeter becomes longer when a naris is collapsed; thus, the aspect ratio should become greater as the perimeter becomes longer.

On the other hand, for the correlation between the visual evaluation and aspect a/u ratio, when the aspect ratio of the naris on both sides became closer, the visual evaluation was higher. Similarly, the correlation between visual evaluation and

Hausdorff distance indicates that when the Hausdorff distance became smaller, visual evaluation was higher. Since the Hausdorff distance is the parameter that indicates how different two given shapes are from each other, it is closely related to the evaluation by the human eye of the symmetry of the two nares. The correlation between the visual evaluation score and Hausdorff distance is shown in Fig. 2.

Five representative cases, including those with the smallest and largest Hausdorff distances are shown in Fig. 3A-E. Fig. 3A. shows the case with the smallest Hausdorff distance, which had a score of 1.01×10^{-3} and a visual evaluation score of 5.00. In Fig. 3B, the Hausdorff distance was 1.71×10^{-3} and visual evaluation score was 3.83. In Fig. 3C, the Hausdorff distance was 2.21×10^{-3} and visual evaluation score was 2.83. In Fig. 3D, the Hausdorff distance was 3.45×10^{-3} and visual evaluation score was 1.67. Fig. 3E is the case with the greatest Hausdorff distance, which had a score of 4.89×10^{-3} and a visual evaluation score of 1.00.

3-2. Evaluation between groups with and without Hotz plate treatment before cheiloplasty.

Next, using these methods, we examined whether there was a difference in left and right naris shape between the group that underwent Hotz plate treatment before the

operation (HP group) and the group that did not (nHP group). There was no difference between the HP and nHP groups in area ratio, perimeter ratio, or aspect a/u ratio. The average area ratio was 0.92 and 0.94 in the HP and nHP group, respectively (Fig. 4A). The average perimeter ratio was 1.00 and 1.02 in the HP and nHP group, respectively (Fig. 4B). The average aspect a/u ratio was 1.18 and 1.16 in the HP and nHP group, respectively (Fig 4C). On the other hand, the score of the visual evaluation tended to be higher in the HP group (HP group: 3.43, nHP group: 2.57, $P = 0.07$, Fig. 4D), whereas the score of the Hausdorff distance was significantly lower in the HP group (HP group: 1.90×10^{-3} , nHP group: 2.87×10^{-3} , $P < 0.0001$). Thus, the visual evaluation score tended to be higher in the HP group, and the Hausdorff distance score was significantly lower (i.e., the similarity of left and right naris shape was significantly higher) in the same group. These findings suggested that using a Hotz plate before the operation will help retain the symmetry of the left and right naris after the operation.

4. DISCUSSION

In this study, for assessing naris symmetry following primary cheiloplasty, we found no significant correlation between visual evaluation and the area ratio or perimeter ratio (correlation coefficient: $r = 0.202$, $P = 0.284$; $r = -0.027$, $P = 0.886$). On the other hand, we found strong correlation between the visual evaluation and Hausdorff distance (correlation coefficient: $r = -0.805$, $P < 0.0001$). There was also correlation between the visual evaluation and a/u ratio (correlation coefficient: $r = -0.47$, $P < 0.001$). These findings indicate that while the aspect ratio, which is the most commonly use measure, correlates with the symmetry assessed by the human eye, the evaluation of similarity by Hausdorff distance correlates even better with human visual assessment. When the Hausdorff distance becomes smaller, the similarity of the nares is higher. Since the Hausdorff distance is used as the algorithm for pattern recognition in computer programs, it is speculated to approximate the way objects are recognized by the human eye.

In comparing the symmetry outcomes between the groups with and without Hotz plate treatment, there was a tendency for the Hotz plate to improve the recovery of shape when judged visually ($P = 0.07$), but not when assessed according to the area ratio, perimeter ratio, or aspect a/u ratio. The only significant difference was detected by the

Hausdorff distance ($P < 0.001$). From these results we can hypothesize that passive orthopedic force of Hotz plate arranged palatal bone properly in horizontal or vertical direction and it supported to improve nasal morphology. In these cases, the same surgeon performed the operation using the same method for 7 years, and started using the Hotz plate in 2004. It is therefore possible that the difference reflected an improvement in the surgeon's skills. In addition finer points of surgical technique in each case may be different. Then the result should be analyzed multi-directionally in further study. Nevertheless, while the Hotz plate group had better naris symmetry visually, of the methods examined, only the Hausdorff distance detected a significant difference, suggesting that the Hausdorff distance method had the most effective power of detection. We found that the Hausdorff distance detected subtle differences that were missed by conventional methods.

Since the facial appearance of patients with cleft lip and palate changes as they grow, including the shape of the nose, it is also difficult to distinguish whether an effect was due to the Hotz plate or to individual physiological factors. To assess the contributions of these factors, it will be necessary to follow the growth for long periods. The Hausdorff distance method allows shape to be evaluated accurately, so it is expected to predict the outcome of a surgical operation more quickly and precisely than

conventional methods. In the future, we plan to use the Hausdorff distance to assess the change in nose shape over time.

In previous studies on evaluating methods for determining naris shape, Lindsay used 10 direct nasal measurements and 3 qualitative visual signs, and Mulliken measured the nasal width and columellar length ^{6,25,26}. It is easy to recognize the left and right symmetry of the nares visually, but it is difficult to evaluate objectively. Nakamura and his colleagues measured the height and width ratio, the ratio of the height of the alar groove, and the curvature of the circle of the nasal ala of the left and right naris ⁸.

Miyamoto and his colleagues imported data using cone beam CT and analyzed the distance and angle from a standard point, which allows shapes to be quantified for analysis and comparison. However, equal values in such analyses do not necessarily mean that two shapes are the same. Anatomical morphology is very complicated and has many fine details, and it cannot be completely resolved simply by increasing the number of standard points. To evaluate surgical outcomes, it is important to establish a method based on human's visual perception. In addition, the devices and software available for measuring and setting landmarks are complicated and labor-intensive. On the other hand, the procedure for taking images of patients' noses for sampling and analysis can also be quite complicated. However, our method of digitizing the similarity

to the control side using photographs with left and right symmetry allows us to assess the degree of similarity with ease.

Other parts of the body, such as the eyes, hands, feet, or ears, can also be digitized in the same manner, and their similarity assessed; thus, our method has the potential for wider application than conventional cheiloplasty assessment methods. In fact, this method can be used for any assessment requiring a judgment of shape, whenever a control shape is available. Moreover, with the development of computer techniques, the availability of 3D data analysis has made it possible to obtain more precise data ^{27,28}.

The Hausdorff distance can also be applied to 3-dimensional shapes, and the software we used in this study can be used for 3D data analysis, as long as there are left and right symmetric 2D or 3D data. In this study, we analyzed 2D data, but in the future, we are planning to compare 3D data. Moreover, we used a combination of commercially available software that is user-friendly. Of note, the postGIS we used for the analysis of geographic information is freeware.

The disadvantage of this method is that the assessment of similarity requires a normal-shaped control side. Thus, to apply it to a two-sided lesion would require modification. For instance, the bilateral cleft lip and palate alar shape is abnormal on both sides. Although it can be measured by Hausdorff distance, it would not be a useful

parameter for the quality of outcome. In these cases, we may need to use an ideal shape as the control, and assess how similar each side is to the control. In this study, to measure the Hausdorff distance, we reversed the images of symmetry around the nose axis, and overlaid the barycenter. Using this method, if the nose is perfectly symmetric, but the positions of the left and right sides of the nose are noticeably shifted, the Hausdorff distance is still 0. In this case, it is necessary to compare the position of the nares as well as their shape. By taking these points into consideration, this method may be improved to make it more useful and broadly applicable.

5. CONCLUSIONS

In this study, we focused on the symmetry of naris shape after primary cheiloplasty of the unilateral cleft lip and palate as an assessment of the quality of the surgical outcome. To determine symmetry, we developed a new method in which we digitized the naris shape and used the Hausdorff distance as a quantitative measure. Our results indicated that the Hausdorff distance detects asymmetry with sensitivity close to visual judgment by the human eye, and digitization enables an objective and accurate analysis. Thus, the Hausdorff distance has the potential to be useful for plastic surgeries in which the results of the operation require symmetry with an unaffected side.

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7. TABLES

Table.1 Cases and each evaluation value

case No.	Hotz plate	side	Visual evaluation	Hausdorff distance ($\times 10^{-3}$)	Area ratio	Perimeter ratio	Aspect a/u ratio
1	Yes	L	3.00	2.28	1.22	1.02	1.10
2	Yes	L	2.00	2.39	1.67	1.25	0.98
3	Yes	R	1.50	2.56	1.01	1.19	1.86
4	Yes	R	2.33	1.83	0.95	1.05	1.18
5	Yes	L	3.67	2.20	0.85	0.95	0.96
6	Yes	L	4.50	1.54	0.98	1.02	1.15
7	Yes	L	2.50	1.71	0.93	0.89	1.36
8	Yes	L	3.83	1.71	1.12	1.01	1.11
9	Yes	L	4.50	1.37	1.37	1.04	1.13
10	Yes	L	4.00	1.16	1.22	1.07	1.23
11	Yes	L	5.00	1.01	1.04	1.04	0.94
12	Yes	R	3.50	2.79	0.81	0.87	1.08
13	Yes	R	1.67	3.45	1.09	1.19	1.90
14	Yes	R	5.00	1.13	0.89	0.91	0.97
15	Yes	L	4.50	1.42	1.00	1.05	0.79
16	No	L	3.00	3.01	0.73	0.79	1.12
17	No	L	3.33	1.78	1.00	1.00	1.02
18	No	L	4.83	1.45	1.15	1.04	1.14
19	No	L	1.17	3.24	1.76	1.18	1.80
20	No	R	1.67	2.16	0.75	0.97	1.87
21	No	R	1.00	4.89	0.32	0.76	1.62
22	No	L	2.83	2.21	0.84	0.84	1.01
23	No	L	1.83	3.04	1.18	0.92	0.92
24	No	R	1.00	4.15	0.64	1.28	1.94
25	No	R	4.00	2.89	1.08	1.03	0.83
26	No	L	1.50	3.51	0.52	0.77	0.67
27	No	L	3.00	3.42	1.25	1.16	0.93
28	No	L	4.00	1.92	0.88	0.89	0.92
29	No	R	1.00	4.02	0.70	0.75	0.56
30	No	L	4.33	1.32	1.08	1.00	1.00

Table.2 Correlation coefficient between each value

	n	correlation coefficient	P	95%CI	95%CI
Visual evaluation: Hausdorff distance	30	-0.805	< 0.0001	-0.903	-0.627
Visual evaluation: Area ratio	30	0.202	0.284	-0.170	0.524
Visual evaluation: Perimeter ratio	30	-0.0272	0.886	-0.384	0.336
Visual evaluation: Aspect a/u ratio	30	-0.470	< 0.01	-0.710	-0.132
Hausdorff distance: Area ratio	30	-0.360	0.051	-0.638	0.000
Hausdorff distance: Perimeter ratio	30	-0.113	0.552	-0.455	0.258
Hausdorff distance: Aspect a/u ratio	30	0.262	0.163	-0.109	0.568
Area ratio: Perimeter ratio	30	0.642	< 0.001	0.367	0.814
Area ratio: Aspect a/u ratio	30	0.00925	0.961	-0.352	0.368
Perimeter ratio: Aspect a/u ratio	30	0.463	< 0.01	0.123	0.706

8. FIGURE LEGENDS

Fig. 1. Measurement of the Hausdorff distance

A: The outline of each naris was plotted using graphic software to extract the shape of the left and right naris.

B: One plot was reversed, saved as an SHP file, in which the center of mass was adjusted with the PostGIS software, to measure the Hausdorff distance.

Fig. 2. Correlation between the Hausdorff distance and visual evaluation.

Plot of Hausdorff distance score versus visual evaluation score. A strong negative correlation was observed between the Hausdorff distance and visual evaluation score ($r = -0.805$).

Fig. 3. Scores of Hausdorff distance and visual evaluation for representative cases.

A: Hausdorff distance score was 1.01×10^{-3} , and visual evaluation score was 5.00.

B: Hausdorff distance score was 1.71×10^{-3} , and visual evaluation score was 3.833.

C: Hausdorff distance score was 2.21×10^{-3} , and visual evaluation score was 2.83.

D: Hausdorff distance score was 3.45×10^{-3} , and visual evaluation score was 1.67.

E: Hausdorff distance score was 4.89×10^{-3} , and visual evaluation score was 1.00.

Fig. 4. Parameters for naris symmetry comparison between the Hotz plate-treated and -untreated groups.

HP group: patients that underwent Hotz plate treatment before the operation. nHP group: patients that did not use a Hotz plate before the operation. N.S: not significant

A: Area ratio. Average 0.92 in HP group and 0.94 in nHP group. N.S.

B: Perimeter ratio. Average 1.00 in HP group and 1.02 in nHP group. N.S.

C: Aspect a/u ratio. Average 1.18 in HP group and 1.16 in nHP group. N.S.

D: Visual evaluation. Average 3.43 in HP group and 2.57 in nHP group ($P < 0.07$). The score in the HP group showed higher trend.

E: Hausdorff distance. Average 1.90×10^{-3} in HP group and 2.87×10^{-3} in nHP group. The score was significantly lower in the HP group ($P < 0.001$).

9. FIGURES

Fig. 1.

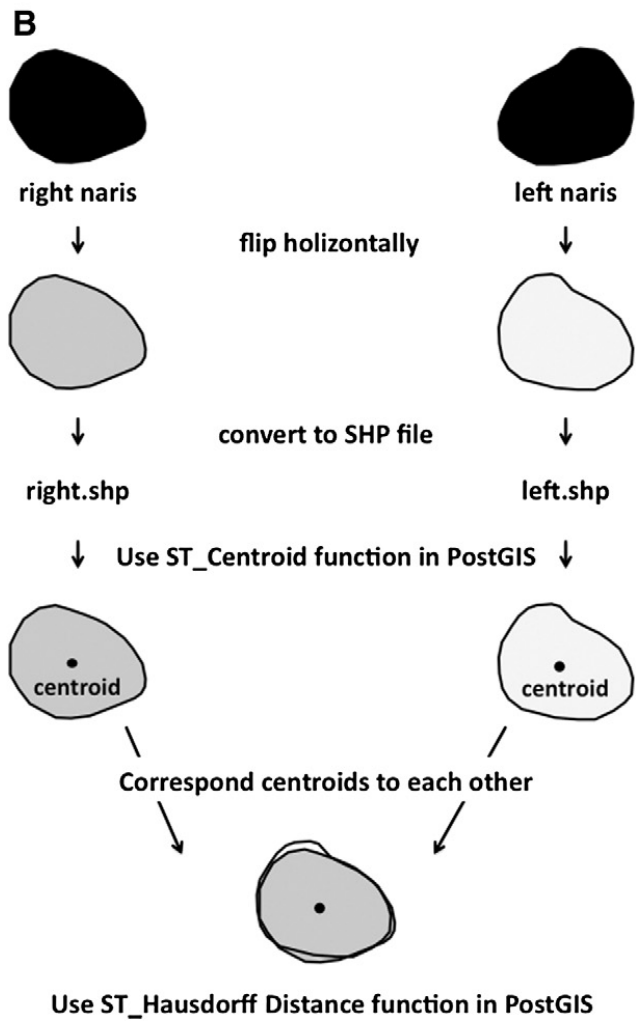


Fig. 2.

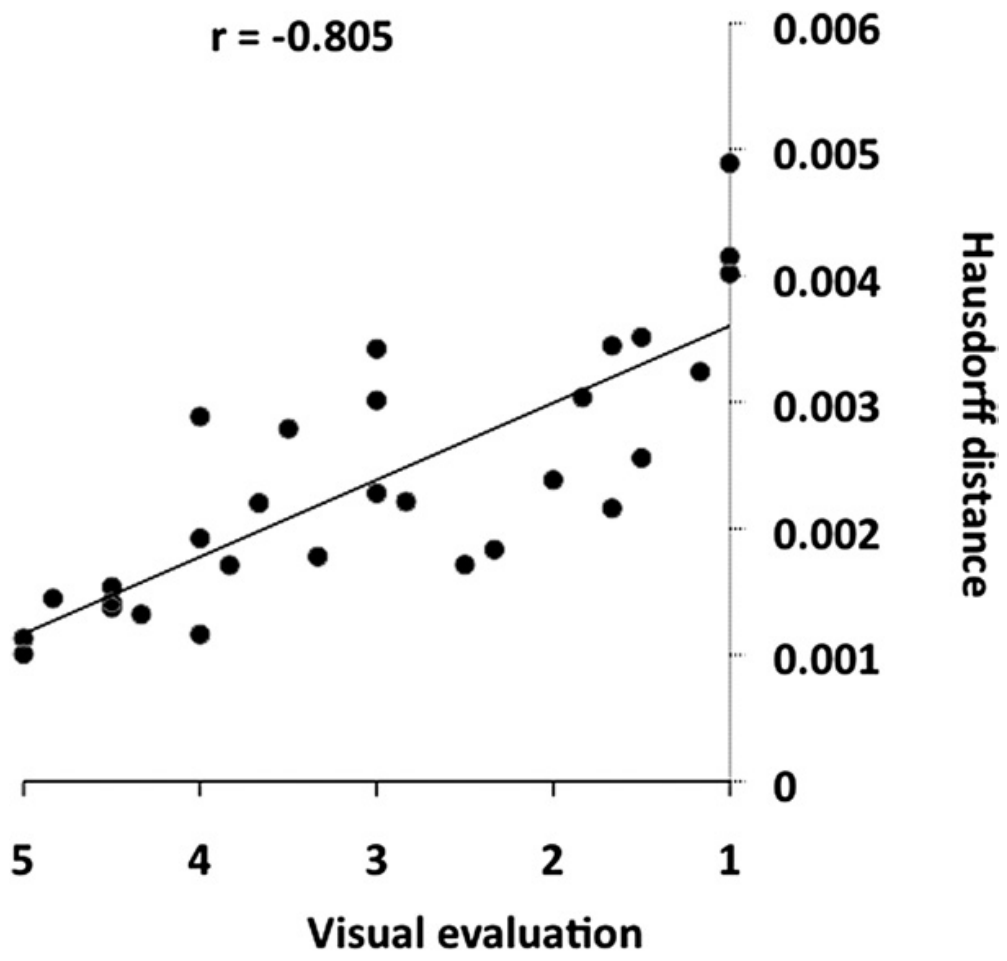


Fig. 3.

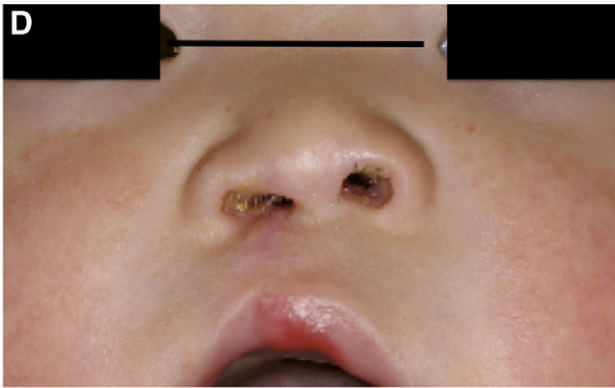
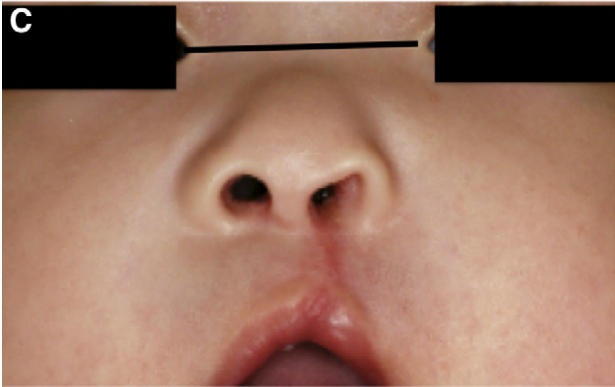
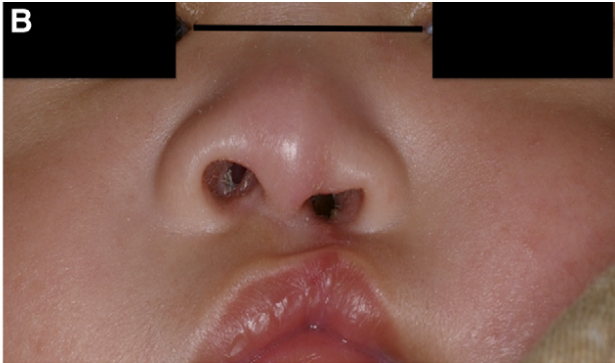
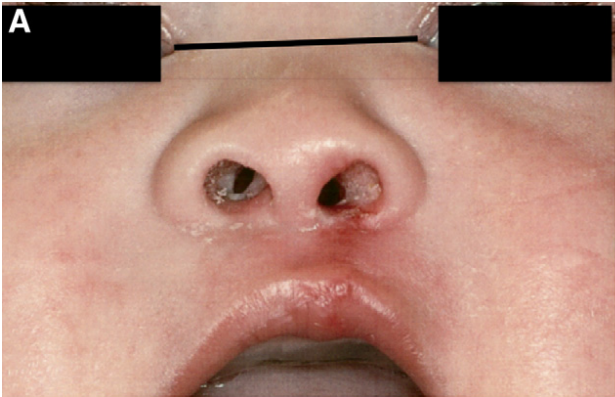


Fig. 4.

