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PŮVODNÍ PRÁCE

Evaluation of aesthetic results of surgical treatment in patients with sagittal craniosynostosis using anthropometry

Hodnotenie estetických výsledkov chirurgickej liečby u pacientov so sagitálnou kraniosynostózou pomocou antropometrie

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SUMMARY

Matejáková L, Horn F, Chrenko R, Štefánková E, Jasenek R, Kuniaková D, Hirošová K, Samohýl M, Mayer-Vargová K, Babjaková J, Jurkovičová J, Argalášová Ľ. Evaluation of aesthetic results of surgical treatment in patients with sagittal craniosynostosis using anthropometry

Objective: The aim of this study is to monitor the cranial parameters of patients with sagittal craniosynostosis. We will mainly focus on the differences between endoscopic and open-surgical treatment and their influence on changes in cranial parameters in patients with craniosynostosis of the sagittal suture. Craniosynostosis has various consequences, including reduced self-confidence and problems fitting in among other children due to craniofacial deformity. Therefore, monitoring aesthetic results of craniosynostosis treatment in pediatric patients is important.

Patients and Methods: The sample of patients included in this study comes from patients monitored by the neurosurgeon at the Department of Paediatric Neurosurgery of the National Institute of Child Diseases in Bratislava, Slovakia. Standardized anthropometric methods and tools to monitor cranial dimensions were used.

Results: In our study, we noted no differences in aesthetic results between open and endoscopic surgical treatment. However, surgical treatment, regardless of the type of chosen treatment, produced the desired aesthetic results, and these results improved over time.

Conclusion: The benefit of the work will be the longitudinal monitoring of the growth of children's cranium, and the evaluation of the effectiveness of surgical treatment in patients with neurocranial growth disorders and the prevention of late consequences of growth disorders.

Key words: craniectomy, craniosynostosis, aesthetic results, anthropometry

SÚHRN

Matejáková L, Horn F, Chrenko R, Štefánková E, Jasenek R, Kuniaková D, Hirošová K, Samohýl M, Mayer-Vargová K, Babjaková J, Jurkovičová J, Argalášová Ľ. Hodnotenie estetických výsledkov chirurgickej liečby u pacientov so sagitálnou kraniosynostózou pomocou antropometrie

Ciele štúdie: Cieľom tejto štúdie je sledovať kraniálne parametre pacientov so sagitálnou kraniosynostózou. Zameriame sa predovšetkým na rozdiely medzi endoskopickou a otvorenou chirurgickou liečbou a ich vplyv na zmeny kraniálnych parametrov u pacientov s kraniosynostózou sagitálneho švu. Kraniosynostóza má rôzne dôsledky, vrátane zníženého sebavedomia a problémov zapadnúť do detského kolektívu v dôsledku kraniofaciálnej deformity. Preto je dôležité sledovanie estetických výsledkov liečby kraniosynostózy u detských pacientov.

Pacienti a metódy: Vzorka pacientov zaradených do tejto štúdie sa skladá z pacientov sledovaných neurochirurgom na Klinike detskej neurochirurgie Národného ústavu detských chorôb v Bratislave. Na sledovanie kraniálnych rozmerov boli použité klasické antropometrické metódy a nástroje.

Výsledky: V našej štúdii sme nezaznamenali žiadne rozdiely v estetických výsledkoch medzi otvorenou a endoskopickou chirurgickou liečbou. Chirurgická liečba však bez ohľadu na typ zvolenej liečby priniesla požadované estetické výsledky a tieto výsledky sa časom zlepšovali.

Záver: Prínosom práce bude longitudinálne sledovanie rastu detskej lebky, zhodnotenie efektivity chirurgickej liečby u pacientov s poruchami rastu neurokránia a prevencia neskorých následkov porúch rastu.

Kľúčové slová: kraniektómia, kraniosynostóza, estetické výsledky, antropometria

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INTRODUCTION

The presence of skull sutures is an integral part of healthy growth and development not only of the skull itself but especially of the child's brain.[1] Each suture or part of it obliterates at a certain age, mostly in adulthood, but if their premature closure occurs in the prenatal or postnatal period, or for their failure to develop, we speak of craniosynostosis.[2,3] In craniosynostosis, we encounter deformation of the skull, because the skull does not grow in the place of the obliterated suture, but parallel to it.[4]

Sagittal suture craniosynostosis is the most common type of craniosynostosis and affects mostly boys. With premature closure of the sagittal suture, the child's skull is so-called boat-shaped (hence the name scaphocephaly or dolichocephaly), it is elongated front-to-back, and the forehead and crown are significantly reduced. A sagittal crest, also called a keel, is often present. It occurs in approximately 2–3 children out of 10,000, which corresponds to about 45% of all craniosynostosis.[5]

In most cases, a patient's craniosynostosis is diagnosed within one year of life. The first step in the diagnosis of craniosynostosis is a thorough examination of either the family history or the history of the patient himself. It is necessary to focus on the occurrence of an atypical skull shape in the patient's relatives, and the exposure of the mother to teratogens and mutagens. Subsequently, the examination of the patient himself is started. The closing of the fontanelles, the protruding edges of the sutures, and the protruding vessels in the subcutaneous tissue of the skull are also observed. As a next step, patient anthropometry is recommended, in which head circumference, head length, and head width are measured, and the *index cephalicus* (IC) is then calculated.[6] A limitation of anthropometry is its inability to capture the overall shape of the skull, as it is only a 2D representation of the skull. For this reason, 3D imaging methods are increasingly used in the diagnosis of craniosynostosis.[7]

Endoscopic craniectomy with the subsequent remodelling of the skull using an orthosis has been used since 1990.[8] In this type of surgical treatment of craniosynostosis, the goal is to remove part or all of the obliterated suture and thereby increase the space in the cranial cavity, allowing the brain to develop properly and reduce intracranial pressure. However, after endoscopic surgery, the shape of the skull changes very slowly and therefore its correction using a remodelling orthosis is necessary. Despite the development of a new endoscopic approach to craniosynostosis operations, open craniectomy is still a frequently used method of surgical treatment. Thanks to a multidisciplinary approach, the overall results and efficiency of open craniectomy have improved in recent decades, and the risk for the patient is also currently reduced to a minimum.[9]

Endoscopic surgery for craniosynostosis is mainly associated with less blood loss during surgery compared to classic, open craniectomy. The advantage of endoscopic surgery is also the reduced hospitalization time of patients compared to open craniectomy. Likewise, the operation time is shorter in the case of endoscopic surgery.[10] Differences in the aesthetic results of surgery in patients undergoing endoscopic craniectomy or open craniectomy have not been found so far. The disadvantage of endoscopic craniectomy is that it can only be performed until a certain age of the patient when the bones of the skull vault are pliable enough to be manipulated with the endoscope. In older patients (from 6 months), proceeding to an open craniectomy is necessary. The advantage of open craniectomy is also the direct remodelling of the skull in the operating theatre, so the patient does not have to undergo therapy with a remodelling orthosis after the surgery. Open craniectomy is also recommended in patients with complex and syndromic craniosynostosis.[11]

In the study, we focused on anthropometric examinations of patients from the National Institute of Child Diseases in Bratislava, Slovakia suffering from sagittal craniosynostosis. Based on the neurosurgeon's recommendation, we monitored patients before surgery and then at defined intervals after surgery and during recovery.

PATIENTS AND METHODS

Patients

Our group consisted of 21 patients with an average age of 17.00 ± 13.24 months at the time of the first examination; four girls (average age at the first examination 21.22 ± 14.45 months) and 17 boys (average age at the first examination 16.65 ± 13.45 months). We performed 39 examinations in total.

In our group, seven patients underwent endoscopic surgery (mean age reached on the day of surgery was 4.48 ± 0.93 months) and 14 patients underwent open surgery (mean age reached on the day of surgery was 13.45 ± 6.12 months). The mean age at which patients underwent surgery was significantly different between patients undergoing endoscopic and open surgery ($p = 0.001$). According to the time that had passed since the surgery at the time of the anthropometric examination, we divided the patients into

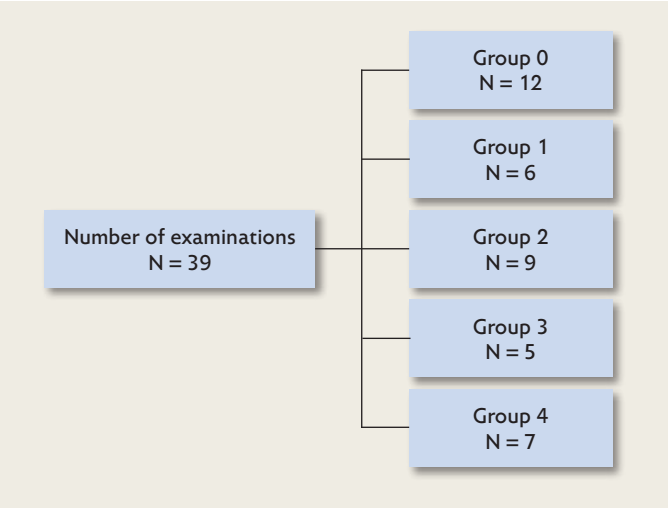

Figure 1: **Number of examinations in each group of patients**

4 groups (Figure 1) – up to one month after the surgery (1), up to six months after the surgery (2), up to one year after the surgery (3) and more than one year after the surgery (4). In these patients, we also performed measurements before surgery (0).

Anthropometric examinations

Part of the anthropometric examinations is the measurement of head circumference, head length, and head width. We proceeded according to the classic methodology of Martin and Saller from 1957[12] and later calculated the cephalic index from these values. We used a metal tape measure to track the circumference of the head, and a spreading caliper to track the length and width of the head.

Definitions of anthropometric cranial points used in data acquisition according to Martin and Saller:[12]

Glabella (g) – is a place between the *arcus superciliares*, above the *sutura frontonasalis*, in the lower part of the *os frontale*.

Opisthocranion (op) – the most dorsal point on the skull in the medial plane. It is most often located in the upper part of the *os occipitale* or on the *protuberantia occipitalis externa*.

Euryon (eu) – the point lying most laterally on the *os parietale* or in the upper part of the *os temporale*.

Definitions of dimensions measured on the skull of patients and indices calculated from these dimensions according to Martin and Saller:[12]

Head circumference (g – op – g) – circumference measured through the glabella and opisthocranion points, in the plane of the greatest length of the head, perpendicular to the medial plane.

Head length (g – op) – direct measurements of the distance between the glabella and opisthocranion points.

Head width (eu – eu) – the greatest width of the head measured perpendicular to the medial plane in euryon points.

Index cephalicus (IC) – defined as the ratio of head width to length multiplied by 100 (%).

$$IC = \frac{eu - eu}{g - op} \times 100$$

IC – *index cephalicus*
eu – eu – head width
g – op – head length

Normalization index (z-score) – after determining the absolute values of the measured dimensions and the relative values of the *index cephalicus*, it is possible to compare these values with the average values of individuals of the same sex and the same age using the normalization index. We obtained these average values from the tables according to Bláha et al.[13] Thanks to this index, we can determine how these values differ from the standard. We calculated it according to the relationship:

$$\text{z-score} = \frac{x - X}{SD}$$

z-score – normalization index
x – the value of the characteristic of the monitored individual
X – average value of the observed characteristic at the same age and sex
SD – standard deviation of the given character in the physiological population

According to the z-score, it is possible to classify the dimensions into categories. Values lower than −2 SD or higher than +2 SD are considered pathological. Values in the range of −1.9 – −1.5 SD are considered very low values but within the variability of the standard. Values in the range of −1.4 – −1.0 SD are considered low values, within the variability of the standard. Values in the range −0.9 – +0.9 SD are average values. Conversely, values of +1.0 – +1.4 SD are defined as high, but within the variability of the standard, and values of +1.5 – +1.9 SD are defined as very high values, within the variability of the standard. The normalization index in the range of ±2 SD represents 95.5% of the occurrence of the distribution of the given character in the physiological population.

Statistical analysis

Obtained data were processed in Microsoft Excel (Microsoft Corporation, One Microsoft Way, Redmond, WA, USA) and in IBM SPSS version 25 (International Business Machines Corp., New Orchard Road, Armonk, NY, USA). We aimed to monitor the results of craniosynostosis treatment and also compare whether the results of two types of surgical treatment (endoscopic craniectomy vs open craniectomy) differ, and if so, which of these two types of surgical treatment is more suitable in terms of aesthetic results.

Table 1: **Comparison of the average values of monitored parameters between groups of patients divided on the basis of the time elapsed since the surgery**

Parameter	0	1	2	3	4	p
HC SD	2.0±1.16	1.18±0.57	1.40±1.00	1.91±0.41	1.09±0.35	0.272
HL SD	2.6±1.17	1.93±0.87	1.71±0.88	2.48±0.18	1.04±0.84	0.121
HW SD	1.8±0.92	1.09±0.88	0.90±0.51	0.53±1.38	0.96±0.76	0.039
Index cephalicus	67.9±3.53	72.86±4.18	74.59±2.19	71.82±0.32	72.41±3.12	0.050

HC – head circumference; HL – head length; HW – head width; SD – standard deviation; 0 – values measured before surgery; 1 – values measured within 1 month after surgery; 2 – values measured within 6 months after surgery; 3 – values measured within 1 year after surgery; 4 – values measured over 1 year after surgery

RESULTS

We noted no differences in the mean absolute values of the normalization index between patients undergoing endoscopic and open surgery. Absolute values of the normalization index of head length ($p = 0.042$; $r = -0.328$) and head width ($p = 0.010$; $r = -0.407$) are significantly, negatively correlated with the time elapsed since the operation. The absolute value of the normalization index of the width of the head and the values of the *index cephalicus* significantly improved with the time that has passed since the surgery (Table 1).

This means that the absolute values of the standard deviation in patients with increasing time since surgery approach zero, the average values of the healthy population. This means that there is an aesthetic improvement in the shape of the skull monitored with the help of anthropometry.

When observing differences in aesthetic results of the surgery, we found no significant differences between endoscopic and open surgery (Figure 2).

In Figure 3 to 6, we can see the development of the absolute values of the patients' normalization index in the period before surgery and during recovery. A significant improvement in these values is visible in all images in our patient cohort. We observe the most significant change in the absolute values of the normalization index of head length, so we can say that the aesthetic results of surgical treatment evaluated by anthropometry are satisfactory.

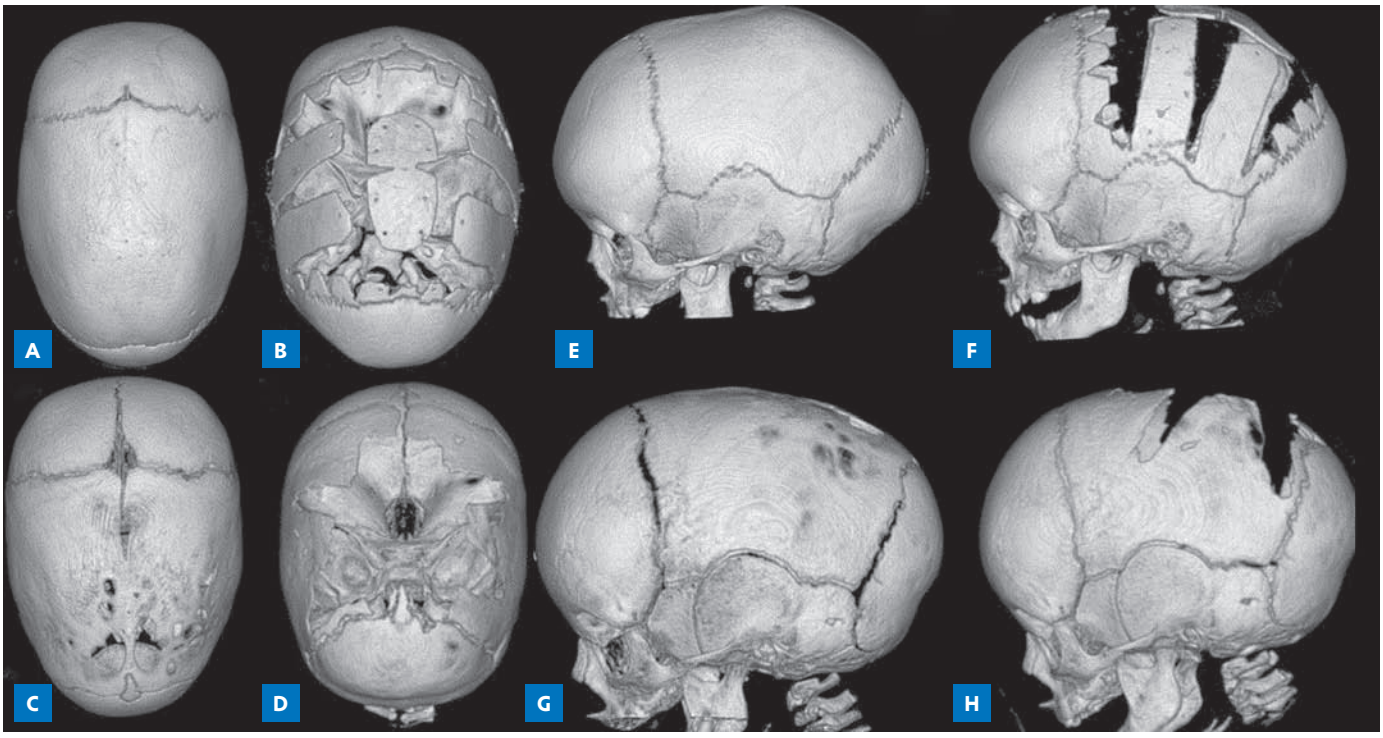

Figure 2: **Comparison of 3D CT images in patients undergoing open (A, B, E, F) and endoscopic surgery (C, D, G, H). A, C, E, G – before surgery, B, D, F, H – after surgery.**[14]

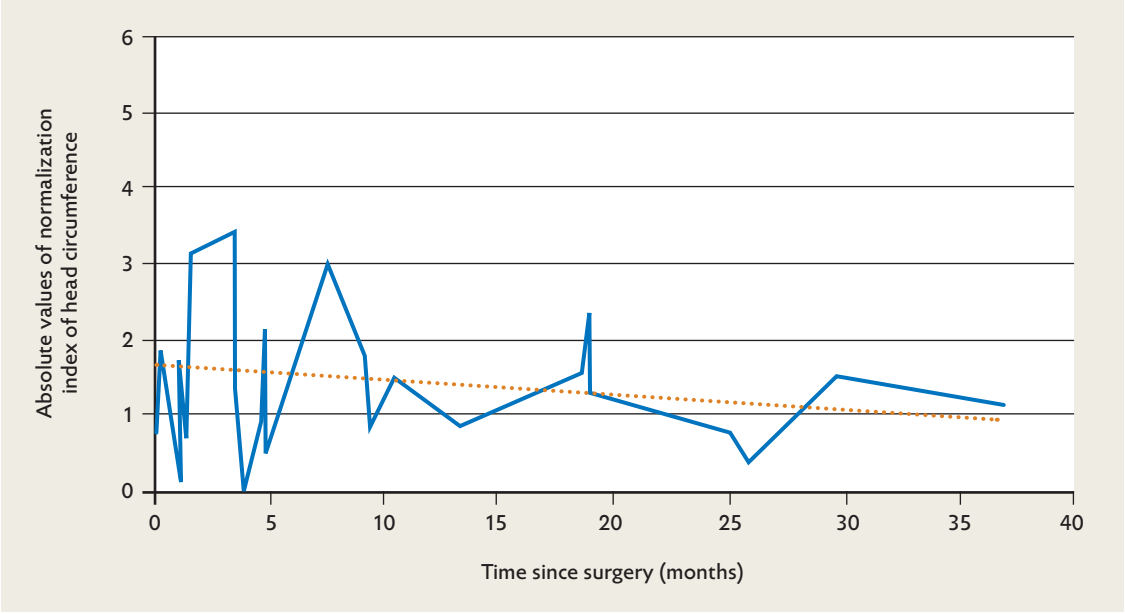

Figure 3: **Development of the absolute values of the standard deviation of the head circumference with respect to the time elapsed since the surgery. The curve shows the changes in these values, and the dashed line shows the trend line.**

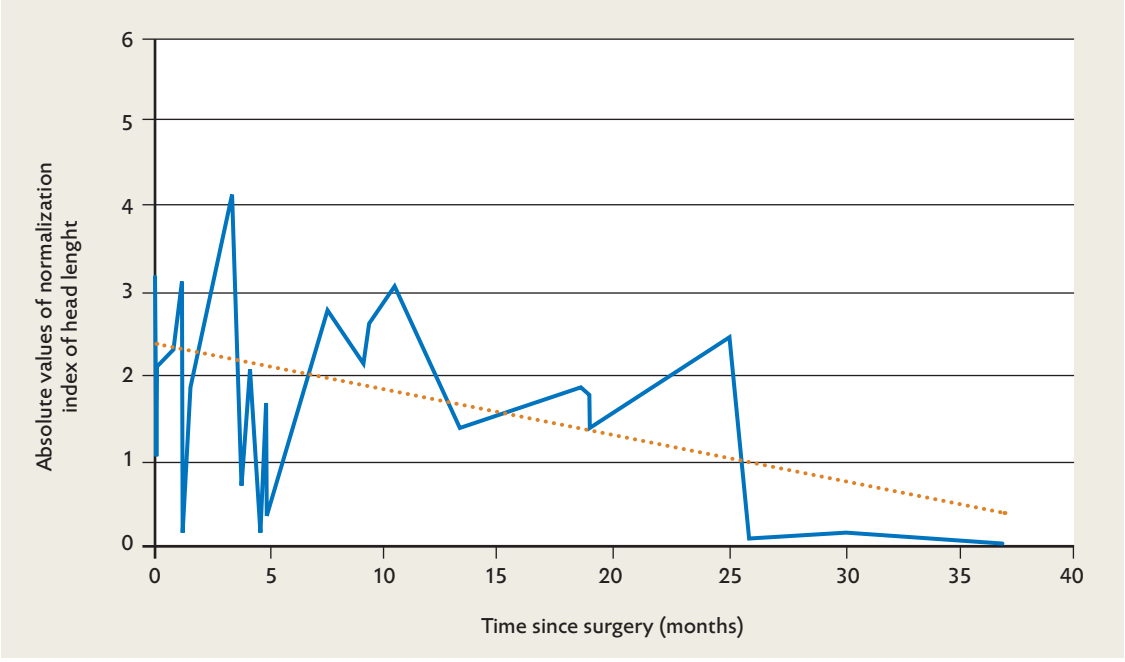

Figure 4: **Development of the absolute values of the standard deviation of the head length with respect to the time elapsed since the surgery. The curve shows the changes in these values, and the dashed line shows the trend line.**

DISCUSSION

Craniosynostosis can have various consequences. The most common consequence is craniocerebral disproportionality and skull deformity, the severity of which is derived from the type of craniosynostosis and the number of obliterated sutures. Another, very serious consequence of craniosynostosis is increased intracranial pressure. The consequences of craniosynostosis affect not only the physical but also the psychological health of the individual. Among other issues, children suffering from craniosynostosis often suffer from low self-esteem and social isolation, mainly due to cranial deformity.[15] Wilbrand et al. noted that the cranial circumference of patients suffering from scaphocephaly is most often above the 90th percentile determined for the given sex and age.[16] Cranial length is above the 90th percentile in nearly 100% of these patients and over 90% is above the 97th percentile, and cranial width is below the 10th percentile in approximately ⅓ of patients with scaphocephaly. However, most of them show average values of skull width. Since the *index cephalicus* is defined as the ratio of head width and length, in the case of scaphocephaly its value is usually lower than the 10th percentile, often below the 3rd percentile.[17]

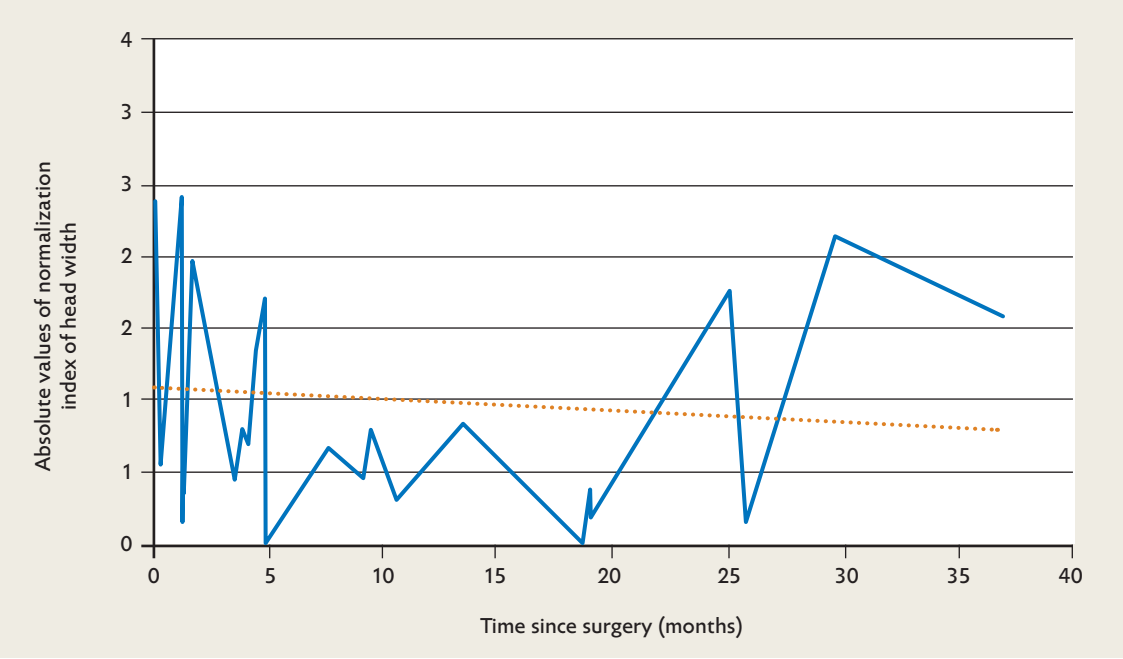

Figure 5: **Development of the absolute values of the standard deviation of the head width with respect to the time elapsed since the surgery. The curve shows the changes in these values, and the dashed line shows the trend line.**

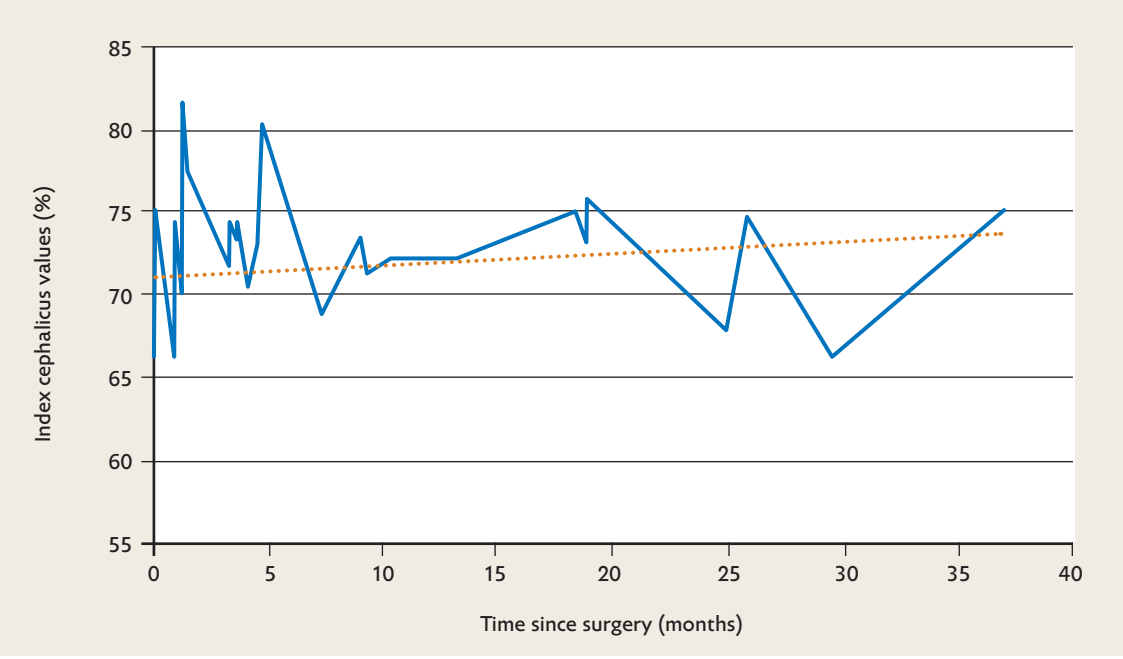

Figure 6: **Development of the *index cephalicus* values with respect to the time elapsed since the surgery. The curve shows the changes in these values, and the dashed line shows the trend line.**

Štefánková et al. observed similar results.[18] In this study, up to 70.0% of patients showed both head length and head circumference values above the 90th percentile, and head length was above the 90th percentile in 21 of 30 patients. A cranial index below the 2nd percentile was recorded in 70.0% of patients. The authors also report an improvement in deviations from the standard and cranial index values after surgery, specifically at the age of 24 months. A statistically significant difference was also noted in the head width values in patients before and after surgery.

The limitation of this study is the small number of patients and that the origin of patients were limited to those from Slovakia. The strength of this research is the collaboration of several clinical anthropologists and pediatric surgeons/neurosurgeons and a complex view of the aesthetics of the child's skull after the difficult surgical treatment of craniosynostosis. It would be important to do a country-wide study in the future.

CONCLUSION

Anthropometry is a suitable non-invasive method, thanks to which we can monitor a pediatric patient for a long time and evaluate the results of treatment. In our sample, we have confirmed the importance of surgical treatment of craniosynostosis from the point of view of the aesthetics of the skull shape. We also found that there are no differences in aesthetic results between endoscopic and open craniectomy in our patients. Our results also brought knowledge about the improvement of the aesthetic results of the surgical treatment of craniosynostosis with the elapsed time since the surgery.

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DATA DEPOSITION

The data that support the findings of this study are available on request from the corresponding author, LM. The data are not publicly available, because they contain information that could compromise the privacy of research participants. |

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