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Original research article

Anthropometry of craniosynostosis

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ABSTRACT

Background: Anthropometry is becoming a popular method for diagnostics of various diseases in pediatric clinical practice. The aim of this study was to assess the growth changes in craniofacial parameters in patients with craniosynostosis and positional plagiocephaly.

Methods: Inclusion criteria for the study were presence of craniostenosis or positional plagiocephaly in a patient with at least three anthropometric evaluations at our department. Studied patients were aged from 1.0 month to 2.5 years with median age at the first and last anthropometric evaluation as 1.83 and 25.27 months, respectively. Further anthropometric results in patients older than 2.5 years were excluded from the study. Statistical significance was tested by the Mann–Whitney test.

Results: The studied group consisted of 70.5% male patients. The type of craniosynostosis was represented by scaphocephaly in 44.1%, by trigonocephaly in 45.6% and by coronal craniosynostosis in 10.3% of the cases. Cranial index was proven as a suitable parameter for evaluating differences in the trend of growth in craniosynostosis ($p < 0.001$) and also for evaluating post-operative results. Significance was found in width of the head ($p = 0.038$) for scaphocephaly and in length of the head for trigonocephaly ($p = 0.001$) in surgically treated patients. Trend of cranial growth in operated patients copied the curve of the norm but in higher or lower values which depends on the type of prematurely closed suture.

Conclusion: Longitudinal anthropometric follow-up is an objective and measurable method that can accurately non-invasively and non-expensively assess skull growth in pediatric patients with cranial deformity.

1. Introduction

Evaluation of head configuration in neonates and infants is very important for early and correct diagnosis of cranial pathologies. Diagnosis is optimally based on physical examination, anthropometrical diagnostic tests and imaging studies.

Cephalometry represents a set of methods for objective assessment of the direct, arc and peripheral dimensions and

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angles of patient's skull. Anthropometric points are specifically defined [9].

Anthropometry is often used in different growth pathologies and underlying skull deformities in patients and the achieved growth parameters are assessed by setting the deviations from the normal physiological growth.

Craniosynostosis (CS) is a premature obliteration of one or more cranial sutures. The cranial growth is trapped in the direction of prematurely fused suture and in other parts of the head the growth accelerates compensatively. Surgical intervention is primarily cosmetic. Anthropometry is a suitable method for evaluating the extent of resulting skull shape deviation. Achieved parameters can also assist the surgeon in decision of indication and timing of surgery. In the long-term period, anthropometry informs about the growth of the cranium and contributes to the assessment of surgery results. Monitoring of anthropometric dimensions in patients in the short- and long-term follow-up optimizes the opportunity to assess the suitability of the treatment method used in patients with craniosynostosis and positional plagiocephaly [13,20].

Most common craniosynostosis, which we analyzed in our study, are described below.

Scaphocephaly – it is a premature synostosis of sagittal seam, with hyperdolichocephalic head shape (Fig. 1a–c). It is the most common case of isolated craniosynostosis, which occurs in 80% of boys. The sagittal suture is the long suture on the top of the head that runs from front to back starting at the anterior fontanel and extending backwards along the middle of the skull to the back of the head. The head typically is elongated in the anterior–posterior diameter and shortened in the biparietal diameter and the ridging of the sagittal suture is palpable. The psychomotoric development is within normal limits in most children [3,10,13,20].

Trigonocephaly – it is a premature synostosis of metopic seam, with the deformity across the frontal and periorbital part of the cranium. Premature fusion of the metopic suture compromises the transverse growth of the forehead which causes a triangular shaped forehead that is associated with

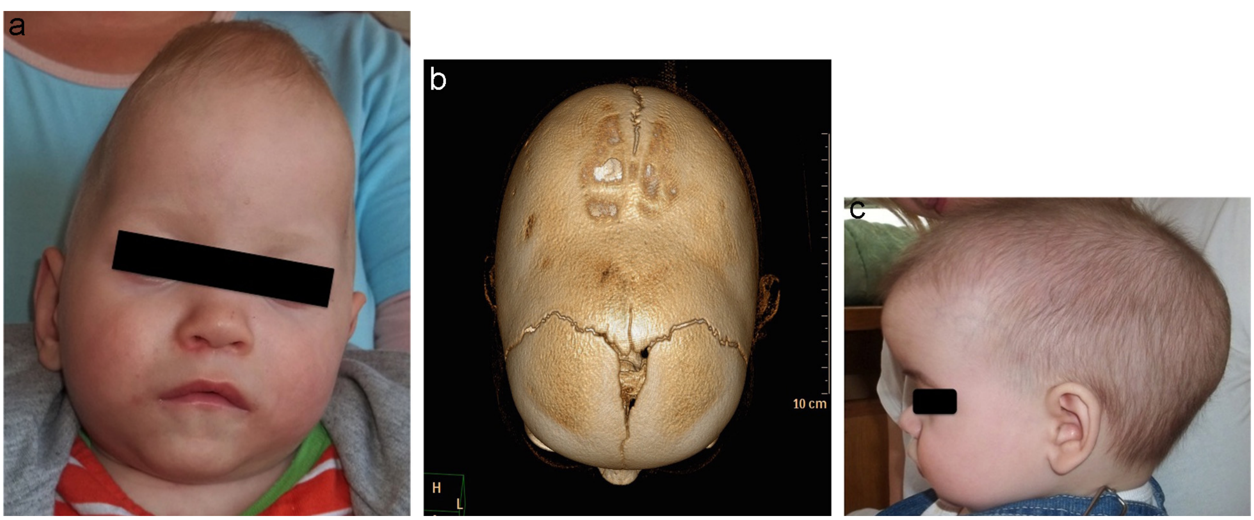

Fig. 1 – (a–c) Scaphocephaly – clinical picture.

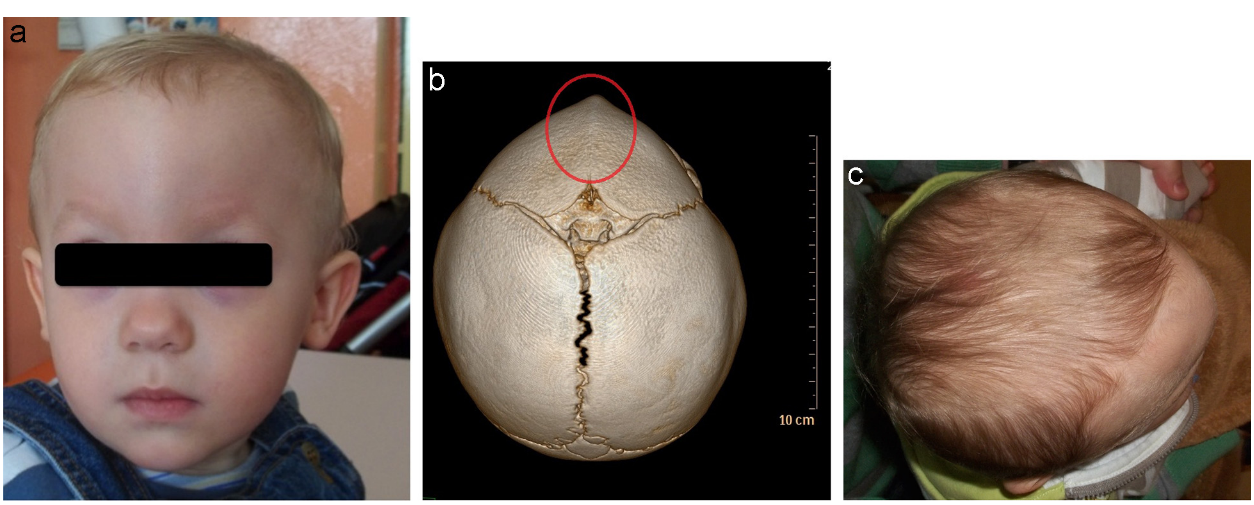

Fig. 2 – (a–c) Trigonocephaly – clinical picture.

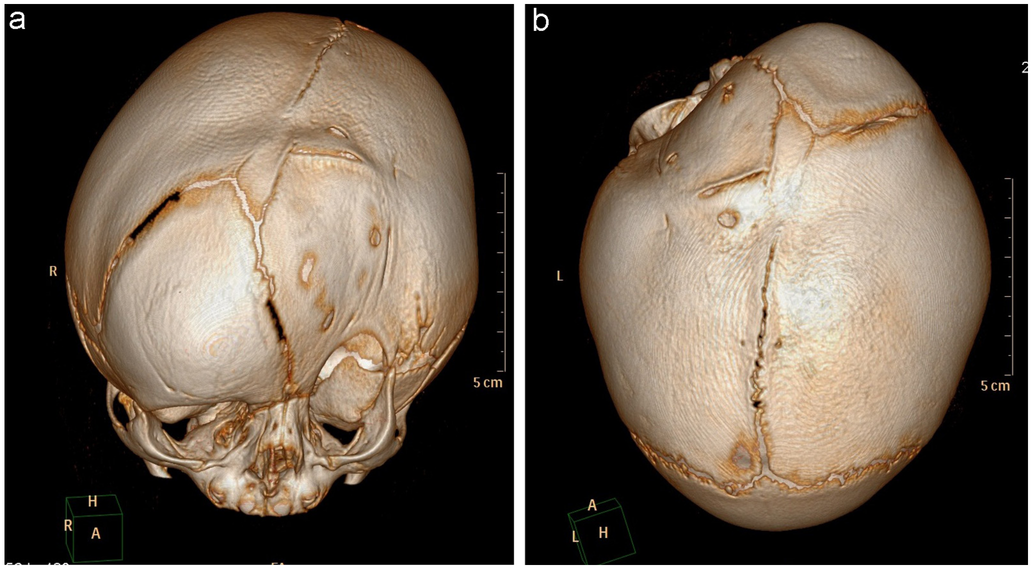

Fig. 3 – (a and b) Plagiocephaly and clinical presentation.

orbital hypotelorism (Fig. 2a–c). Hypotelorism occurs as a result of underdevelopment of the ethmoidal bone, contrary to compensatory rise of the bi-temporal size of the skull. The deformity can vary from mild to severe. There is usually a prominent mid-frontal ridge down the forehead that can be seen or felt and the eyebrows may appear ''pinched'' on either side. The eyes are usually spaced closer to each other than normal, causing a definite recognizable deformity of the forehead and eyes [3,10,13,20].

Plagiocephaly – it is a unilateral synostosis of lambdoid seam or coronal seam (Fig. 3a,b). The resulting deformity does not only affect the skull, but it also results in underdevelopment of the ipsilateral frontal bone (forehead), supraorbital ridge (eyebrow) and anterior cranial fossa, giving a flattened appearance. The pathological changes also extend to involve the base of the skull resulting in lower facial deformity. True lambdoid synostosis (posterior plagiocephaly) is a very rare form of craniosynostosis and is commonly mistaken for positional plagiocephaly. In true lambdoid synostosis, the lambdoid suture is fused. This gives the affected side a flattened appearance along the back of the head and when looking down at the patient, the ear on the affected side is pulled back toward the involved suture. The forehead is usually not affected as severely but may appear flattened.

In contrast, with positional plagiocephaly (Fig. 4a–c), the backside of the head is flat but the ear is pushed forward and the forehead on that same side appears full. Additionally, in deformational plagiocephaly, the skull shape abnormality may not have been present at birth and may improve over time, while in posterior plagiocephaly, due to the true synostosis of the lambdoid suture, the deformity is present at birth and worsens with time. Torticollis, prematurity and cervical spine abnormalities are often suggestive of posterior positional molding [3,10,13,20].

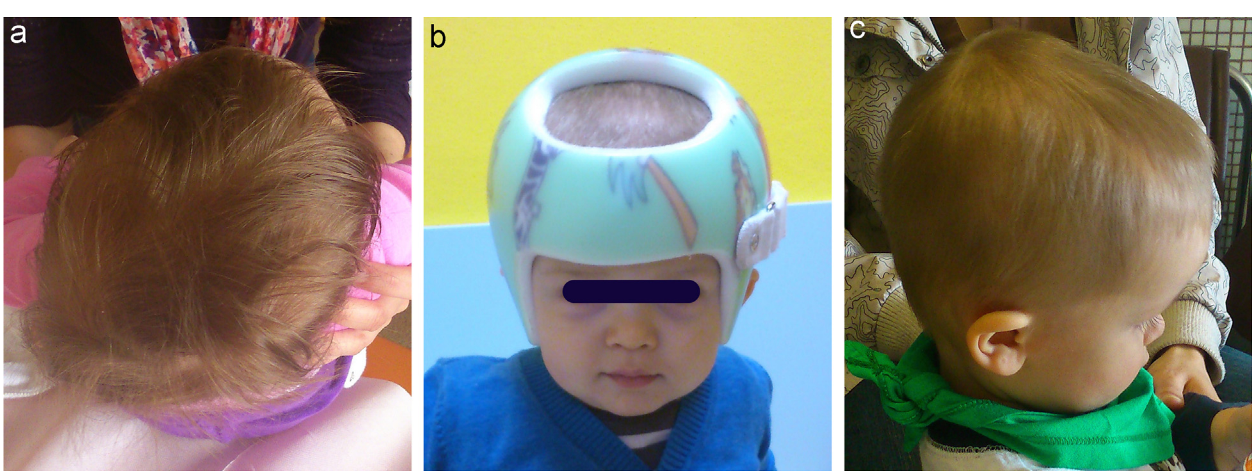

Fig. 4 – (a–c) Positional plagiocephaly.

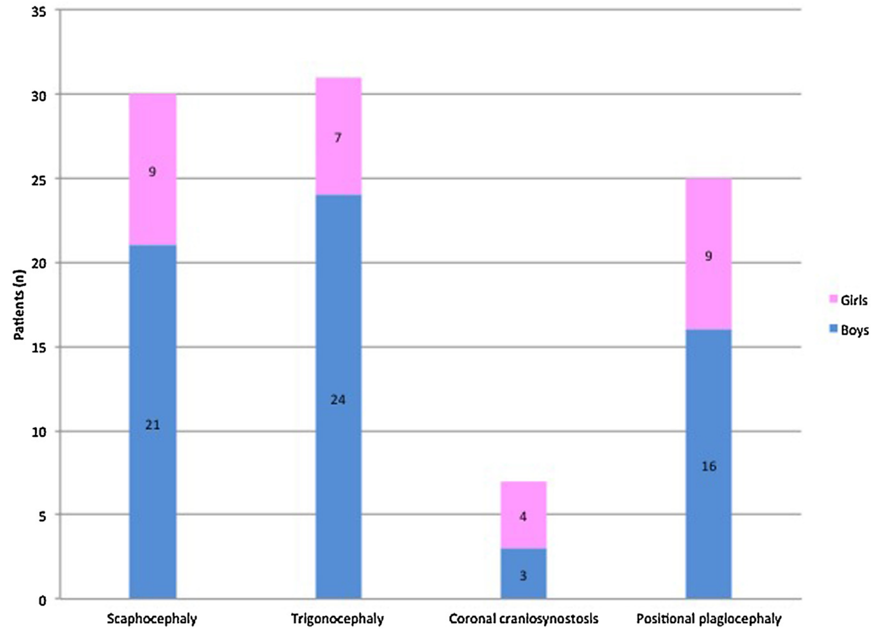

Fig. 5 – Group of analyzed patients.

2. Patients and methods

Patients for statistical analysis were chosen based on their diagnosis – our study included all patients with craniosynostosis and positional plagiocephaly aged from 1.0 month to 2.5 years, with median age at first examination as 1.83 months and at last examination 25.27 months. We excluded patients who underwent 2 and less anthropometric evaluations and patients older than 2.5 years. Patients with cranial deformity were monitored longitudinally. All measurements were carried out at the Department of Paediatric Surgery Children's University Hospital in Bratislava. We analyzed all acquired anthropometric dates of patients meeting the inclusion criteria in the period between October 2010 and November 2014. Ethics committee approved the participation in the study.

Study included 68 patients with craniosynostosis (Fig. 5) divided into three groups according to the obliterated suture and 25 patients with positional plagiocephaly.

The objective of our study was to evaluate the impact of the diseases on craniofacial parameters and assessment of the growth trend of cranium with respect to the applied surgical intervention.

3. Anthropometric measurement

Anthropometric measurement provides the opportunity to document the children's craniofacial parameters and cranium shape at the beginning, during and at the end of the treatment. Important craniofacial dimensions are: the head circumference, the maximum head length, the maximum head width, the minimum width of the forehead, the face width, the morphological height of the face and the width of the skull base (Fig. 6a,b). Measurements are performed by standard anthropological technique according to Martin and Saller [11] and its modifications. Instruments required for anthropometric measurement are the spreading calliper, the sliding calliper and the measuring tape.

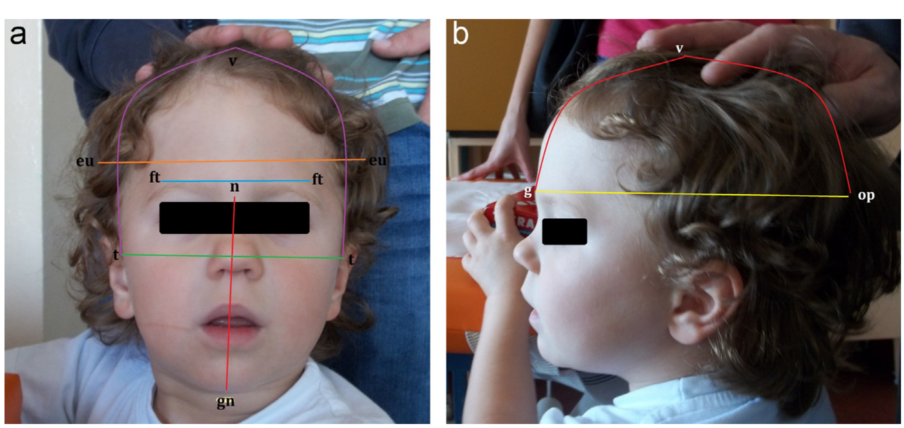

Fig. 6 – (a and b) Anthropometric measurement.

The anthropometric evaluation in patients with craniosynostosis is carried out as the first clinical examination of the child (ideally new born). Subsequently, further anthropometric evaluations are scheduled (depending on the type and severity of craniosynostosis) until the surgery – once a month, every 3 months and before surgery. Measurements are carried out also after surgical intervention and during the recovery period (once a month, every 3 months, every 6 months, once a year). For some patients surgical treatment is primarily cosmetic therefore not all the patients with CS are operated on (based on the discourse with parents and extent of fusion of the suture involved). The patients with CS without surgery are monitored long-term by anthropometry at the same intervals as the children who were operated on.

The history of sleeping position in infancy is a very important fact in differential diagnosis, because positional anomalies of the cranium are states that can imitate craniosynostosis. It could be non-syndromic plagiocephaly, physiological dolichocephaly or physiological brachycephaly [10,16]. Anthropometry can differentiate the primary craniosynostosis and the positional plagiocephaly. Moreover, the anthropometric monitoring of the patient can document the trend of cranial growth (pathological or not) and provides mosaic examinations under which the surgeon indicates or does not indicate the intervention.

4. Evaluation

The evaluation of anthropometric dates can be performed in various ways. The fastest way to determine the head shape is using indexes. The most commonly used index, which shows the relationship between the width and length of the head is called cranial index (index ICE), which is calculated as width of the head × 100/head length (Table 1). Configuration of neonatal skull is most commonly mesocephalic head shape and may vary in the early postnatal period. In the past, brachycephaly was the most prevalent (Fig. 7). Dolichocephaly used to be rare in the past but now is more common [14,22].

Another specific index that is used in differential diagnostic of skull pathologies is the diagonal index. Diagonal index is mostly applied in the evaluation of positional anomalies – plagiocephaly, as remodeling helmets specifically treat the severe forms of these deformities. The patient is assessed before the helmet treatment, during recovery and after the treatment is completed [16]. Based on this index we determine the type and severity of plagiocephaly in a patient. An index value less than 3.5% is considered to be without deformity, whereas diagonal values of the index greater than 13.5% indicate severe deformity. On the basis of the observed values, cranial and diagonal indexes can be used for determining the percentage of degree of asymmetry.

Table 1 – Categories of cranial index [6].

Categories of cranial index	Boys	Girls
Hyperdolichocephal	x–70.9	x–71.9
Dolichocephal	71.0–75.9	72.0–76.9
Mesocephal	76.0–80.9	77.0–81.9
Brachycephal	81.0–85.4	82.0–86.4
Hyperbrachycephal	85.5–90.9	86.5–91.9
Ultrabrachycephal	91.0–x	92.0–x

Another way to evaluate and observe the measured anthropometric parameters is the evaluation by normalization index. The absolute values of the dimensions and the relative values of the indices of patients with cranial deformities are compared with the physiological average values and the indices of the whole body. We use norms of physiological population from Bláha et al. [2] and Slováková et al. [19]. The normalized ratio is used to convey a proportional analysis of the body, allowing the comparison of any number of anthropometric parameters of different gender and ages as well as statistical evaluation of the mean difference from the norm [4,17].

We use growth percentile graphs for a detailed analysis of the cranial growth. They demonstrate the pathology of the anthropometrical parameters and also the average growth curve of the norm. We focus on changes in the cranial index in scaphocephaly, trigonocephaly and positional plagiocephaly. Percentile graphs are compiled on the basis of standards [2].

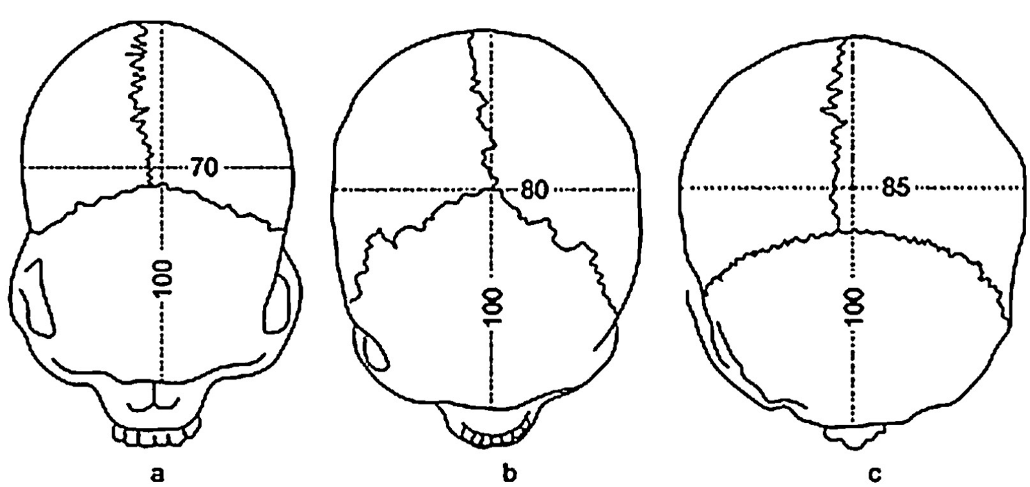

Fig. 7 – Average head shapes [7].

5. Results

Here we present the most important results of anthropometric evaluation in patients with craniosynostosis and positional plagiocephaly.

1. *Scaphocephaly* ($n = 30$): it is premature synostosis of sagittal suture, with hyperdolichocephalic head shape. It is the most common type of isolated craniosynostosis, which we observed in our studied group in 44.11% of patients ($n = 68$). Scaphocephaly affected 70% male population. There is a slight male preponderance according to International Society for Paediatric Neurosurgery.

The *craniofacial dimensions*, which indicate the possible pathology, are the head circumference (HC) and maximum head length (Lmax). We observed deviations above the 90th percentile from the norm in 21 patients and 70.0% of patients for HC and Lmax, respectively. The *HC median value* in the 3rd month of patients' life was 43.1 cm (90th percentile) in comparison with the norm which was 41.2 cm. Median value of HC after surgery (in the 24th month of patients' life) was 50.0 cm (50th–75th percentile). Value of norm was 49.2 cm (50th percentile). *The Lmax value* in the 3rd month of patients' life was 15.0 cm (90th percentile) in comparison with the norm which was 13.8 cm (50th percentil). After surgery the median value of Lmax was 17.8 cm in the 24th month of patients' life (75th–90th percentile). Value of norm was 16.8 cm (50th percentile). The most common categories of cranial index were dolichocephaly and hyperdolichocephaly.

Trend of growth in cranial index

The graph (Fig. 8) shows that before surgical treatment, cranium has the tendency to grow faster lengthwise (we observed cranial index under 2 percentile in 70.0% of patients) and after surgical treatment the growth of the width renews with subsequent change of cranial index from hyperdolichocephalic to dolichocephalic head shape. Therefore, in the 24th month of patients' life dolichocephaly became prevalent in patients with operated scaphocehaly compared to the standard population where brachycephaly or mesocephaly is dominant.

We observed significant enlargement of width of the head after surgical treatment ($p = 0.038$). The aim of the operation is to slowdown the growth acceleration in length and renewal of the growth in width, which we confirmed by anthropometry and by statistical significance.

2. *Trigonocephaly* ($n = 31$): in our study we observed 31 patients with trigonocephaly, with 77.4% of the male population. Important *craniofacial dimensions* are minimum width of forehead (Fmin) and maximum head length (Lmax). The monitored deviations from the norm ranged in minimum width of the forehead below 10 percentile (77.4% and 24 patients, respectively). When analyzing the cranial index, hyperbrachycephaly, ultrabrachycephaly and brachycephaly are prevalent. In non-operated patients ($n = 18$) we found similar values as in operated patients. In these non-operated cases mild form of trigonocephaly prevailed.

The *Fmin median value* in the 3rd month of patients' life (non-operated) was 7.65 cm (10th percentile) in comparison with the norm which was 7.9 cm (50th percentile). Patients who were operated had in their 3rd month of life the Fmin median value 6.8 cm (10th percentile). Median value of Fmin after surgery (in the 24th month of patients' life) was 8.8 cm (50th–25th percentile). Value of norm was 9.1 cm (50th percentile). These two groups of patients did not differ in value of Fmin ($p = 0.63$).

The Lmax value in the 3rd month of patients life (non-operated) was 13.6 cm (50th–25th percentile) in comparison with norm which was 13.8 cm (50th percentile). Patients who were operated had in their 3rd month of life the Lmax median value 14.2 cm (50th–25th percentile). Median value of Lmax after surgery (in the 24th month of patients' life) was 16.10 cm (50th–25th percentile). Value of norm was 16.8 cm (50th percentile).

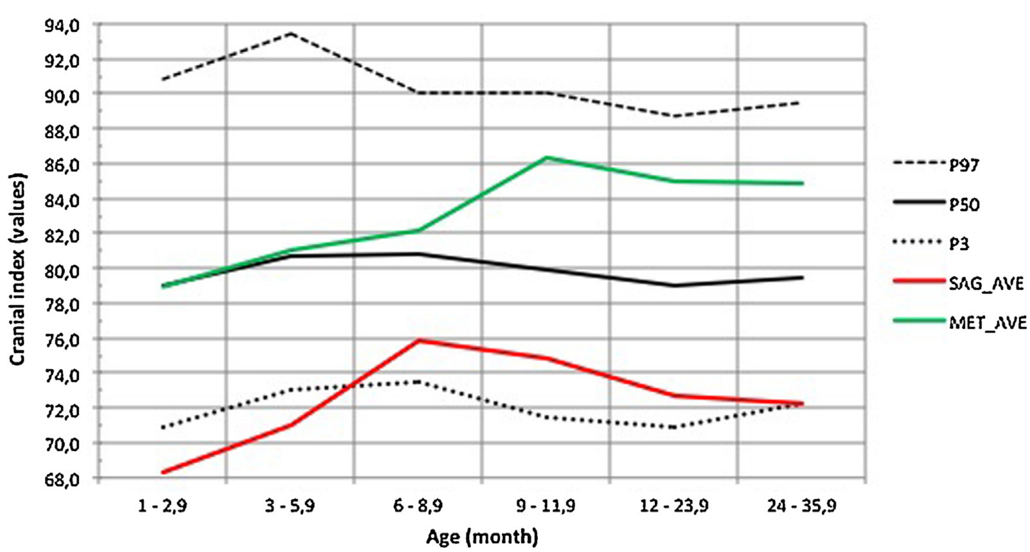

Fig. 8 – Trend of growth in scaphocephaly and trigonocephaly (cranial index). SAG_AVE – average trend of growth curve in scaphocephaly; MET_AVE – average trend of growth curve in trigonocephaly; P97, P50, P3 – percentiles and average trend growth curve of normal population.

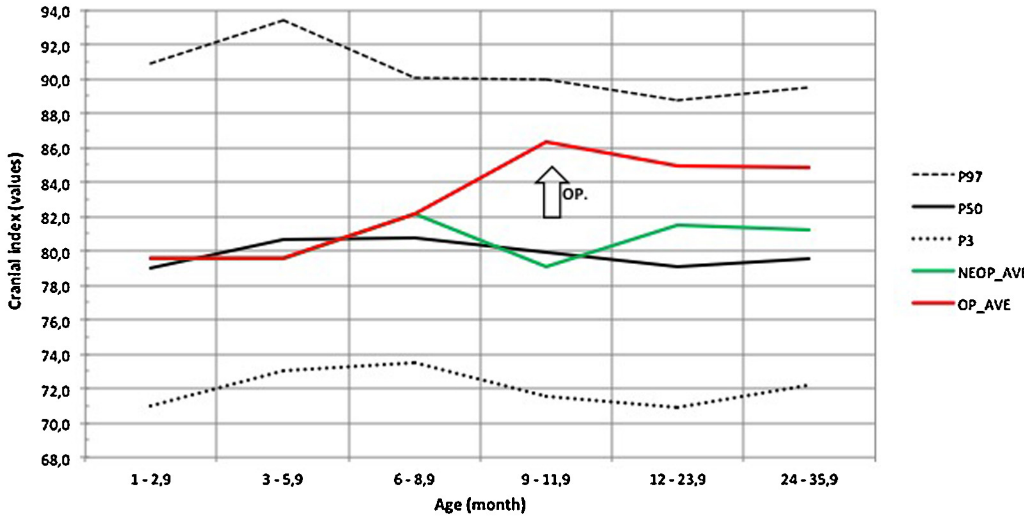

Fig. 9 – Trend of growth in trigonocephaly (cranial index)-operated and non-operated group. NEOP_AVE. – average trend of growth curve without surgery; OP_AVE. – average trend of growth curve with surgery; P97, P50, P3 – percentiles and average trend growth curve of normal population.

Trend of growth in cranial index

The ratio of the width/length of the head copies the growth curve of the norm to the 7th month of patients life in both groups (operated and non-operated). In the group of operated patients, there prevailed higher values of the width and hyperbrachycephaly that dominated in 37.5% and 9 patients, respectively. In recovery period (after surgery) the growth curve divided, in the non-operated group prevailed brachycephalic head shape in 62.5% of patients. The growth trend of cephalic index remains below the line of the healthy population but copies the normal growth curve (Fig. 9).

Significant differences in cranial index between scaphocephaly and trigonocephaly were found in categories of cranial index ($p < 0.001$). These two types of craniosynostosis have different clinical and also anthropometrical characteristics, which we approved (Fig. 8).

We have not found a significant difference in values of cranial index in the first year of life between the group of patients with trigonocephaly who were operated on and the group who were not operated on ($p = 0.24$). This fact is explained by a milder form of trigonocephaly in a group of non-operated patients.

We found significance ($p = 0.001$) in the length parameter in operated patients, so it means that the surgical intervention positively affected on this length parameter, which copies the norm growth curve in postoperative period.

3. *Positional plagiocephaly ($n = 25$):* in these cases we assessed the trend of growth especially in cranial index.

Trend of growth in cranial index

Cranial index for evaluation of this type of deformity is not reliable and we must use also other indexes for exact analyses. The range of growth varies considerably and also depends on the extent of asymmetry. Another important parameter is the length of the head, which is below average (Fig. 10), as the result of positional anomaly. We also compared a group of patients ($n = 5$) who were treated by remodeling helmet. We assessed that the value of cranial index copied the norm growth curve after treatment by helmet. For statistical significance, it is necessary to measure more patients (Fig. 10).

6. Discussion

Anthropometric measurement is one of the objectively non-invasively measurable diagnostic methods in pediatric patients with cranial deformity. Using the anthropometry, the cosmetic effect on patient's cranium can also be evaluated. Early recognition of premature synostosis of one or more sutures ensures the surgical observation and longitudinal anthropometric monitoring (comparison with the standard and evaluation of the deviation from the norm of the parameters, growth acceleration or deceleration, etc.). Based on our analysis we can predict trend of growth in each type of cranial deformity and we can also discourse with parents how surgical intervention can help in this type of diagnosis. Craniofacial measurements are helpful in the decision-making for a surgeon to operate and to choose which type of intervention is most suitable for the patient.

Wilbrand et al. [25] evaluated anthropometric data via percentile based assessment of CS. They focused to find an objective method to assess cranial deformation based on normative craniofacial percentiles. Matching of anthropometric data of CS patients with craniofacial norms could be useful in grading the clinical picture and potentially adapting the operative procedure.

In complex CS could be analyzed through the foramen magnum (FM). The FM area is significantly altered in FGFR3-related brachycephaly. The study confirms the importance of FGFRs on FM growth whereas TWIST-1 seems to have a minor role [5].

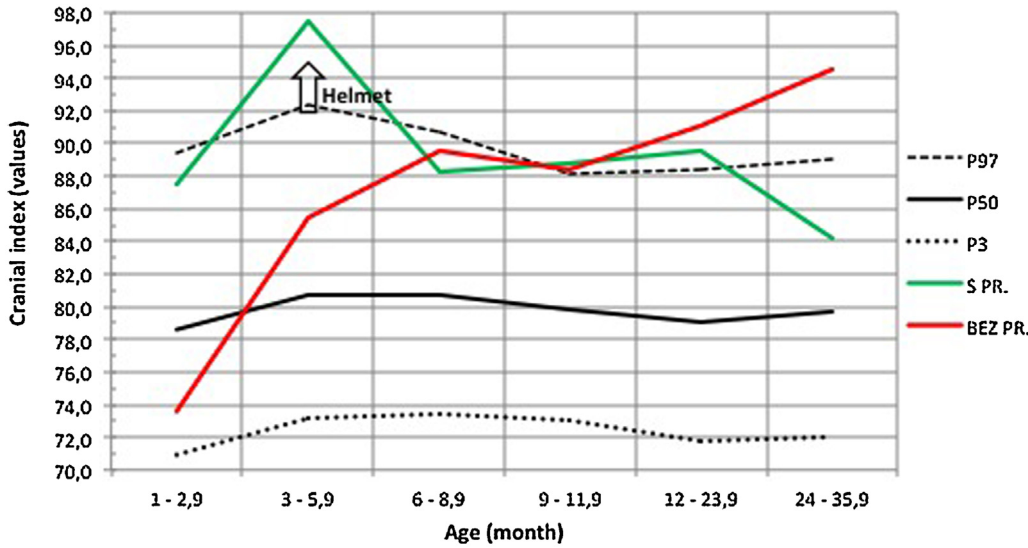

Fig. 10 – Trend of growth in positional plagiocephaly – with and without helmet (cranial index). S PR. – average trend of growth curve with helmet; BEZ PR. – average trend of growth curve without helmet; P97, P50, P3 – percentiles and average trend growth curve of normal population.

The clinical course is variable and influenced by multiple factors, acting at different steps of the child's growth. Intracranial hypertension is a major concern already in the first month of life; active cerebrospinal fluid dynamics disorders, venous hypertension, and progressive craniocerebral disproportion are considered the main pathogenetic factors [21].

Surgical intervention in our opinion is ideally placed at the age of 6–8 months of life while the bone is more plastic, easily remodeled and we observe a better result in head shape.

Other research groups have mentioned these results of surgical treatment. Surgical procedures, which aim at expanding the posterior cranial vault, have come to play an increasingly important role in the treatment of syndromic CS. The posterior cranial vault may be expanded by formal cranioplasty or by less invasive methods based on gradual posterior cranial vault expansion or distraction. Data of these authors [15] suggest that these three techniques offer a potential for a significant increase in skull volume.

In another retrospective case-controlled study authors introduce a comparative study of anterior cranial vault distraction versus remodeling. Anthropometry (length, height, circumference, volume) revealed no significant differences between the two treatment groups [8].

In our group of patients we found some important significances in anthropometrical parameters.

Significant differences were confirmed in the operated group of patients on the basis of craniofacial parameters. We observed significant enlargement (in scaphocephaly) of width of the head after surgical treatment ($p = 0.038$).

Wilbrand et al. [25] confirmed that most children with sagittal synostosis were above the 90th percentile for cranial circumference and length, whereas only 27.9% were below the 10th percentile for cranial width.

The team of authors [12] collected serial anthropometric data of children before and after total cranial vault remodeling and evaluated cranial vault growth pattern. They found significant differences in long-term cranial growth pattern and relapse into dolichocephal skull growth.

In trigonocephaly we found significance ($p = 0.001$) in the length parameter in operated patients, so it means that the surgical intervention positively affected on this length parameter, which copies the norm growth curve in postoperative period.

For trigonocephaly, Wilbrand et al. [25] found normal cranial circumference values in most patients (10th–90th percentile), 40.9% were above the 90th percentile for cranial length and 63.1% and 57.9% were above the 90th percentiles for sagittal and transverse circumferences.

Metopic synostosis can be divided into two distinct severity indices according to Beckett et al. [1]. They found that the severe group has significantly narrower orbitofrontal dimensions, whereas the moderate group does not differ from control.

The authors Seruya et al. [18] compared cranial growth across three patterns of fronto-orbital remodeling for metopic synostosis. Retrocoronal patterns provide long-term gains in head circumference percentile and the least growth impairment in cranial length. Irrespective of osteotomy design, expansion in frontal breadth relapses significantly over time.

Significant differences in cranial index between scaphocephaly and trigonocephaly were found in categories of cranial index ($p < 0.001$). These two types of craniosynostosis have different clinical and also anthropometrical characteristics, which we approved. The aim of the operation is to slowdown the growth acceleration in length and renewal of the growth in width, which we confirmed by anthropometry and by statistical significance (growth depends of type of obliterated suture). We did not find significant difference in values of cranial index in the first year of life between the group of patients with trigonocephaly who were operated on and the group who were not operated on ($p = 0.24$). This fact is explained by a milder form of trigonocephaly in a group of non-operated patients.

Also other authors use cranial indices in diagnostic process of CS. Wilbrand et al. [25] found that most (83%) children with scaphocephaly had cranial indices below the 10th percentile.

Van Lindert et al. [24] focused on validation of cephalic index measurements in different ways – manually using a caliper, using skull X-ray, 2D CT and 3D CT images and with help of plagiocephalometry. The conclusions are that significantly different results are achieved when using 2D CT relative to the manual caliper determination. No significant differences are observed between the 3D techniques and the manual method.

The important role of anthropometry is also in differential diagnostics. In primary craniosynostosis–plagiocephaly one parameter stops growing or grows more slowly (depending on the type of prematurely fused seam). Therefore anthropometry has crucial importance, in such cases, in the diagnosis without other methodologies (MRI or CT scans).

Anterior plagiocephaly represents the most challenging simple suture craniosynostosis. The clinical differential diagnosis with other forms of cranial asymmetry is possible on the grounds of mere clinical findings. A classification system is necessary not only for establishment of surgical planning but also to predict the late cosmetic and functional outcomes [6].

The main conclusion of Netherlands authors about helmet therapy was that they discourage the use of helmet as a standard treatment for healthy infants with moderate to severe skull deformation. Their theory is based on the equal effectiveness of helmet therapy and skull deformation following its natural course, high prevalence of side effects and high costs associated with helmet therapy [23].

7. Conclusion

Monitoring of the patient's anthropometric dimensions in the short- and long-term period provides the optimal opportunity to assess the cranial growth and the suitability of the methodology that was used in patients with craniosynostosis and positional plagiocephaly. Non-invasive anthropometry is an inevitable part of diagnostics, surgical treatment indication, postoperative follow-up and evaluation of the outcome of the treatment.

8. Conflict of Interest

None declared.

9. Financial disclosure

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10. Ethics

The experiments comply with current laws of Slovak republic.

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