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The Cephalic Index Is Not Different Among Groups of Children Aged 36 to 48 months with Chronic Otitis Media with Effusion, Recurrent Acute Otitis Media and Controls

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Abstract

Objective—The Cephalic Index, an anthropometric measure of head shape, was reported to be different between individuals with and without signs of past or concurrent otitis media (OM). In this study, we compared the Cephalic Index and other measures of head shape among groups of children aged 36 to 48 months with a documented history of chronic OM with effusion (COME), recurrent acute OM (RAOM) and CONTROLS (few to no OM episodes) to test that hypothesis.

Methods—In 41 CONTROL, 36 COME and 42 RAOM children, Maximum Head Width, Maximum Head Length and Head Circumference were measured and the Cephalic Index (Head Width/Head Length X 100) was calculated. The four measures were compared among the three groups using a General Linear Model that included group, sex and race as factors and age as a covariate.

Results—There were no differences among groups in the Cephalic Index or Head Length. Head Width was significantly smaller in the RAOM when compared to the CONTROL group and Head Circumference was significantly smaller in the RAOM and COME groups when compared to the CONTROL group. For all measures, the distribution of values showed significant overlap among groups.

Conclusions—These results do not support the hypothesis that the Cephalic Index is different between young children with and without OM, but did document differences in Head Width and Circumference among groups. However, the large overlap in each measure for the three groups suggests that none capture sufficient information on Eustachian tube anatomy to predict disease presentation.

Keywords

Cephalic Index; Anthropometry; Otitis Media; Head Shape

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CONFLICT OF INTEREST STATEMENT

None of the authors have a conflict of interest with respect to the materials presented in this manuscript.

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INTRODUCTION

Past studies relate inefficient Eustachian tube (ET) function to a predisposition for the development and persistence of otitis media (OM) [1]. During growth and development in infants and children, there are measureable changes in the size and orientation of the ET system (ET, paratubal muscles) that are associated with improved ET function [2]. Because the various components of the ET system are intimately related to the craniofacial skeleton, the extant morphology of that complex is believed to affect ET function and, thus, OM susceptibility, and some past studies support this relationship [3–7]. Of interest in this regard are the results published in the German literature by Pautow (cited by Worley and colleagues [8], Stolovitzky and Todd [9]) who measured the head shape of 50 adult cadavers and made casts of the ETs [10]. He reported that the Cephalic Index, a measure of head shape calculated as the Maximum Head Width divided by the Maximum Head Length multiplied by 100, was related to ET morphology. Specifically, a high Cephalic Index indicative of round headedness (brachycephaly) was associated with a relatively straight ET and poorly defined ET isthmus, a low Cephalic Index indicative of long headedness (dolichocephaly) was associated with a sharply angled ET and a narrow isthmus, and an intermediate Cephalic Index (mesocephaly) was associated with a moderate ET angulation and a well-developed isthmus. These results were interpreted as suggesting that a brachycephalic (and perhaps a dolichocephalic) head shape predisposes to aberrant ET function and, by consequence, OM.

Two previous studies explored the possible relationship between the Cephalic Index and OM prevalence. Stolovitzky and Todd studied 198 adolescent and adult male subjects aged 9 to 75 years [9]. They measured the Maximal Head Length and Head Width to calculate the Cephalic Index and examined the tympanic membranes using photoendoscopy. They classified the individuals as having bilaterally normal tympanic membranes ($n=95$), “at least one abnormal” tympanic membrane ($n=13$) or indeterminate with respect to tympanic membrane state ($n=79$). When dichotomized to normal and “at least one abnormal” tympanic membrane, a trend was noted for the former group to have more dolichocephalic head shapes when compared to the more brachycephalic head shapes in those with “at least one abnormal” tympanic membrane. The average Cephalic Index over all age groups was significantly less for those with normal (76.5 ± 2.5) when compared to those with abnormal tympanic membranes (78.8 ± 2.6) and was intermediate (77.3 ± 3.8) for those with an indeterminate tympanic membrane rating. Problems with this study are the wide range of ages included in the sample, the low number of persons with “abnormal tympanic membranes”, the failure to define the specifics of what constituted an “abnormal tympanic membrane” and the use of simple statistical tests to evaluate the significance of comparisons despite the wide range of ages (racial distribution not reported).

In the second study, Worley and colleagues enrolled 203 otherwise normal children who were 4 or 5 years of age (127 White, 76 Black; 92 Male) [8]. They measured the Maximum Head Length and Head Width of all subjects to calculate the Cephalic Index and performed bilateral tympanometry. Type B tympanograms were taken as evidence of concurrent middle ear disease (OM point prevalence equaled 23%). To analyze the data, they defined brachycephalic head shapes as those with a Cephalic Index greater than the average for the population by 1 standard deviation, dolichocephalic head shapes as those with a Cephalic Index less than that for the population by 1 standard deviation and mesocephalic head shapes as those with a Cephalic Index included within the two bounds. The mean Cephalic Index for the population was 76.1 ± 3.7 . Overall, there was a significant difference in the frequency of type B tympanograms by head shape with more brachycephalic children presenting that abnormality when compared to mesocephalic children. Problems with this study are the failure to account for demographic factors in the formal analysis, the arbitrary

assignment of the three head shapes, and the fact that the diagnosed OM may have been a transient and not a constitutive condition resulting from, for example, a recent viral upper respiratory tract infection.

In the current study, we identified three groups of otherwise healthy children between 36 and 48 months of age presenting to our research clinic and subsequently enrolled in a long-term follow-up of the changes in ET form and function and OM prevalence with age: those with a history of chronic OM with effusion (COME), those with a history of recurrent acute OM (RAOM) and those with no significant history of OM (CONTROL). Each diagnosis was confirmed by a review of medical records. All children included in this study had anthropometric measurements done. Using these data, we tested the primary hypothesis that the Cephalic Index is different among the three groups. The limited age range, well characterized OM history and presentation, and use of statistical procedures that accounted for the contribution of demographic factors to head shape avoid many of the problems associated with the two earlier studies discussed above.

MATERIAL AND METHODS

In this study, we included 119 children, 36–48 months of age who had no significant history of OM (CONTROL: N=41; Male=20; White=29, Black=12; average age = 3.6 ± 0.3 years), a history of COME (N=36; Male=21; White=30, Black=6; average age = 3.6 ± 0.3 years) or a history of RAOM (N=42; Male=20; White=33, Black=9; average age = 3.4 ± 0.3 years). The parents of all children signed an institutionally approved Informed Consent for their child's participation.

The children were classified as having no significant history of OM if they had not had tympanostomy tubes (TTs) inserted and did not satisfy the criteria for a positive history of COME or RAOM. RAOM was defined as having 3 or more episodes of symptomatic OM in one year or 5 or more episodes by study entry, with at least 2 episodes in the previous year. COME was defined as having had 3 or more consecutive months of middle ear effusion (MEE) if bilateral, 6 consecutive months of MEE if unilateral or 3 or more episodes of OM lasting for at least 2 months each with at least one episode of OM in the year prior to entry. Children may or may not have had patent tubes or a tympanic membrane perforation at the time of evaluation. Children who had TTs were classified according to the reason for surgery. All diagnoses for these children were confirmed by review of their medical charts.

The following measures were made using standard techniques and anthropometric calipers/tapes: Maximum Head Circumference, Maximum Head Length and Maximum Head Width [11]. Specifically, Maximum Head Circumference was measured as the length of the arc passing from ophryon (the mid-plane of a line tangent to the upper limits of the eyebrows) to opithocranium (the most prominent posterior point of the occiput) and returning to ophryon. Head Length was measured as the distance between glabella (the most prominent point in the median sagittal plane between the supraorbital ridges) and opithocranium (the most prominent posterior point of the occiput). Maximum Head Width was measured as the distance between the bilateral euryons (the most lateral points of the head in the parietal region) [12]. The reported coefficients of variation for repeated measures of these variables in 3-year old children are 0.3, 0.4 and 0.3, respectively [13]. The Cephalic Index was calculated as the Maximum Head Width divided by the Maximal Head Length multiplied by 100 [11]. We analyzed the data for each measure using a General Linear Model (Analysis of Covariance) that included disease group, gender and race as factors and age as a covariate. Because this procedure measures the significance of the difference in the age-adjusted variable among the 3 groups, *post hoc* paired significance testing was done using the Tukey-Kramer Multiple-Comparison Test when applicable. The data presented by Stolovitzky and

Todd [9] show that a sample size of 28/group would have approximately 80% power to detect a true difference in the Cranial Index between the control and a diseased at the typical alpha of .05 for between group comparisons. Thus, for the main outcome variable, this sample size for our study is sufficiently powered. We also adopted the procedure described by Worley and colleagues in their publication on head shape and OM [8]. Specifically, the average and standard deviation of the Cephalic Index for the entire group was calculated and head shape was defined as brachycephalic if the Cephalic Index exceeded the average plus 1 standard deviation, dolichocephalic if the index was less than the average minus 1 standard deviation and mesocephalic if lying within those bounds. For each disease group, the number of children assigned to each of these head shapes was evaluated for statistical significance using a 3×3 contingency test.

Because past studies compared the Cephalic Index between groups defined as “no disease” and “disease”, we repeated these analyses for 2×2 comparisons of the data for the CONTROL group (“no disease”) versus the combined COME and RAOM groups (“disease”). All statistical tests were done using the NCSS 97 Statistical Program (Kaysville, Utah).

RESULTS

The Table reports the average and standard error of the age-adjusted cephalometric variables for each group, the significance level of any difference among groups, and the *post-hoc*, pairwise comparisons that were significant as determined by the ANACOVA. Contrary to our hypothesis, there was no significant difference among groups in the Cephalic Index. Also, there was no significant difference among groups in Head Length, but Head Width and Head Circumference were both significantly different among the 3 groups. *Post-hoc*, pairwise comparisons showed that the CONTROL vs RAOM difference was statistically significant ($p < 0.01$) for Head Length and both the CONTROL vs RAOM ($P < 0.01$) and CONTROL vs OME comparisons ($P = 0.04$) were statistically different for Head Circumference. For these comparisons, the CONTROL average value was greater than the average value for the comparison group. These analyses also documented a significant sex-by-group interaction for the Cephalic Index ($p = .02$) and a significant effect of sex on group assignment for Head Length ($p = .01$) and Head Circumference ($p < 0.01$), but no effect of race on any variable. When repeating these analyses for groups defined as CONTROL and OM (RAOM plus COME), again, only Head Width ($p = 0.05$) and Head Circumference ($p < 0.01$) were significantly different among groups.

Using the head shape definitions developed by Worley and colleagues [8], the number of brachycephalic, dolichocephalic and mesocephalic individuals was 6 (15%), 4 (10%) and 31 (76%) for the CONTROL group; 5 (14%), 6 (17%) and 25 (69%) for the COME group, and 6 (14%), 7 (17%) and 29 (69%) for the RAOM group. There were no differences in the distribution of head shapes among these 3 groups ($\chi^2 = 1.06$, $p = 0.90$). We also explored other methods to assign the head shape from the Cephalic Index such as using the standard error rather than standard deviation or using tertiles to make the discriminations, but in all cases, there was no relationship between head shape and group assignment. When the groups were reassigned as CONTROL and OM (RAOM and COME), a 2×3 contingency analysis showed no significant between-group difference in the distribution of head shapes as defined by Worley and colleagues ($\chi^2 = 1.06$, $p = 0.59$).

To determine if any of these measures capture information that can be used to assign a given child to his/her respective group, for each measure and group, we determined the number and percent of observations that lay outside of the bounded range defined by the average plus or minus one standard deviation of the CONTROL group. The respective percentages

for the CONTROL, OME and RAOM groups were 27%, 28% and 24% for the Cephalic Index ($\chi^2=0.18$, $p=0.92$), 29%, 33% and 29% for Maximum Head Width ($\chi^2=0.24$, $p=0.89$), 34%, 39% and 31% for Maximum Head Length ($\chi^2=0.54$, $p=0.76$), and 24%, 29% and 23% for Head Circumference ($\chi^2=0.38$, $p=0.82$). None of the four measures discriminated groups based on this criterion.

DISCUSSION

A causal relationship between craniofacial anatomy, ET structure, ET function and OM susceptibility has been suggested based on previous work [2]. In infants and children, the timed activity of the various growth regions of the craniofacial skeleton and the synchronization across regions creates an evolving scaffold from which the ET system is suspended, grows and matures into its adult form [14]. Abnormal growth or synchronization patterns during development are expected to adversely affect the anatomy and function of the ET system and, consequently, predispose to the different OM expressions. In that regard, past studies have shown differences between OM and control groups in nasopharyngeal size [5], facial morphology [3, 6, 14, 15], the relative length and angle of the components of the cranial base [14, 15], the bony portion of the ET [4, 16] and the projections of the ET and associated musculature onto the lateral, parasagittal plane [14], among others.

However, attempts to synthesize these results are fraught with difficulties associated with study design and measuring technique. For example, the main tool previously used to assess these relationships, cephalometry, does not directly image the ET system, and thus many of the proposed effects of craniofacial development on the ET system are inferential, at best. Newer techniques that are capable of imaging the ET system such as Computerized Tomography are being applied, but have as yet failed to identify craniofacial markers in adults that assign an individual's susceptibility to the designated OM expression under study [17]. A second difficulty is that these studies evaluated children over wide age ranges as well as adults, where different craniofacial growth patterns are operative or have been completed and different ET-ME system morphologies are extant. A third difficulty is that the studies failure to discriminate different OM expressions such as uncomplicated acute OM, OM with effusion, chronic suppurative OM, RAOM and COME, or else focused on only one of the expressions, though the underlying ET function is expected to be different for each expression or combination of expressions.

Nonetheless, these types of studies are important for two reasons. Mechanistically, they may provide convincing evidence to confirm the expected relationship between the morphology of the ET system and the susceptibility to a specific OM expression, thus satisfying the form-function-disease paradigm. Categorically, these studies may identify certain craniofacial characteristics in those young children who are likely to develop a particular OM expression thus allowing for close follow-up and monitoring of the "at risk" children and/or they may identify craniofacial characteristics of those individuals with extant OM disease expressions who are more or less likely to resolve their condition without aggressive management.

In that regard, anthropometry offers a number of significant advantages when compared to the other techniques utilized to describe the craniofacial complex. These include the relative ease of the measurements, the lack of a requirement for expensive and sophisticated instrumentation and the lack of exposure of the subject to ionizing radiation. Balancing these advantages is the disadvantage associated with an inability to capture important details with respect to internal structures such as the ET and cranial base. For this reason, the past observation that the Cephalic Index, a simple anthropometric measure, is different in older children and adults with and without a marker of past/present OM (abnormal tympanic

membranes) and is different in younger children 4 and 5 years old with and without tympanometric evidence of extant OM (Type B tympanogram) is both intriguing and bears further study. As outlined in the Introduction, those studies suffer from a number of shortcomings that were partially addressed in the current study. Specifically, we confined the study population to a relatively homogenous group of otherwise normal children aged 36 to 48 months of age, the specific disease phenotype was well documented by chart review, and the statistical methods included provisions to control for race, sex and age. However, our results did not support a difference in the Cephalic Index among children with RAO, COME or CONTROLS. Also, using the analysis advocated by Worley and colleagues and variations on that theme [8], the defined head shapes did not discriminate the subjects with respect to disease group membership. Interestingly, Head Width and Head Circumference were different among groups, with significant pairwise differences between the CONTROL (larger) and RAO groups for the former and between the CONTROL (larger) and both the RAO and OME groups for the latter. To our knowledge, this has not been previously reported but may present an alternative measure of head shape to the Cephalic Index for purposes of defining groups based on OM presentation. However, because of the large overlap of all measures among groups, we consider it to be unlikely that Head Width or Head Circumference will prove useful in assigning a given individual to a particular disease group. Nonetheless, we plan to follow-up on this observation during our ongoing studies of the growth and development of the ET system and to reanalyze all anthropometric data for these groups of subjects at later ages.

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TABLE 1

TABLE A Summary of the Primary Results for the Analysis of Covariance that included Group Assignment, Sex and Race as Factors and Age as a Covariate. Listed Entries are the Age-Adjusted Average (AVG) and Standard Error (SE) of the Anthropometric Measurements (in mm) for Each of the Three Groups, the Probability (P) Value for the Comparison Among the Three Groups and the Post-Hoc Significance of Paired Comparisons (where applicable)/

VARIABLE	CONTROL (N=41)		COME (N=36)		RAOM (N=42)		P-Value
	AVG	SE	AVG	SE	AVG	SE	
Head Width	137.0	0.9	135.9	1.0	133.0	0.9	0.04 *
Head Length	172.8	1.1	172.9	1.2	171.5	1.1	0.72
Cephalic Index	79.4	0.7	78.8	0.8	77.5	0.7	0.27
Head Circumference	511.1	2.9	502.5	3.1	497.3	2.9	0.02 **

/ The effects of sex, race and interactions on these variables is presented in the Text of the Results.

* Pairwise RAOM vs CONTROL significant at $P < 0.01$ by the Tukey-Kramer Multiple-Comparison Test.

** Pairwise OME vs CONTROL significant at $P = 0.04$, RAOM vs CONTROL significant at $P < 0.01$ by the Tukey-Kramer Multiple-Comparison Test.