The effects of Perceived Severity of Skull Deformation on the Treatment Preference of Youth Health Care Physicians and Paediatricians in the Netherlands

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Abstract

Goal: The goal of this study is to determine which factor of the perceived severity of skull deformation is most defining and in what manner this perceived severity of skull deformation is influencing the treatment preferences of youth health care physicians (YHCP) and paediatricians. Design: Clinical vignettes were used in a conjoint analysis setting where both respondent groups gave their opinion on severity and treatment preference. Each question contained a set of attributes (Oblique Diameter Difference Index, Cranio Proportional Index, Occipital Lift, Ear Deviation and Gender) which were depicted as stylized deformed heads. The vignettes were accompanied by a questionnaire on experience with plagiocephaly and its treatments. Results: The results from the multivariate regression analysis showed that the ODDI is the largest contributing factor with a β=0.367 ($P<0.05$) for YHCP and β=0.461 ($P<0.05$) for Paediatricians in the Severity. This model for Severity with ODDI, CPI, OL, ED and Gender had a goodness-of-fit of R^2_a=0.392 for YCHPs and R^2_a=0.298 Paediatricians. The severity was also a major factor in the formation of a treatment preference, correlating with treatment preference with a R=0.717 for YHCPs and R=0.633 for Paediatricians. Considering that additional variables contribute to the treatment preference, for example the physicians personal preference resulted in an R=0.243 for YHCP and R=0.434 for the Paediatricians. This combined model had a goodness-of-fit of R^2_a=0.572 for YHCP and R^2_a=0.505 for Paediatricians. Conclusion: It is unclear if more levels of OL would have made a difference in this model. The Severity is likely a modifier for the personal treatment preference a physician might have. This would explain the overall gravitation to the 'wait-and-see' treatment in both the respondent groups. Additional factors for severity and treatment preference should be explored. The influence of additional variables on Severity, Treatment Preference and subsequent Advice given to the parents was not explored in this study but would be crucial in understanding the general decision-making process a physician has to go though when dealing with plagiocephaly.

Keywords: Plagiocephaly, Skull Deformation, Paediatricians, Youth Health Care Physicians, Clinical Vignettes, Conjoint Analysis, Treatment Preference, Severity Assessment, Oblique Diameter Difference (ODDI), Cranio Proportional Index (CPI), Occipital Lift, Ear Deviation, and Gender.

Introduction

In 1992 the American Academy of Pediatrics (AAP) issued a recommendation on sleeping positions to reduce the number of Sudden Infant Death Syndrome (SIDS) in new born infants [1]. The (American) National Institute of Child Health and Human Development (HICHD) initiated the 'Back to Sleep'-campaign in 1994 which resulted in a drop in SIDS

incidences over the following years [2]. In 2006 the number of reported SIDS incidents was at 0.55 deaths per 1000 live births, compared to 1.4 per 1000 in 1988 [3]. Coincidentally after the start of the Back to Sleep-campaign, an increase in head deformities like plagiocephaly and brachycephaly was reported [4], Skull (or Head) Deformation (SD) is the overarching term used to describe the medical conditions of Plagiocephaly (which is a slanting of the cranium), Brachycephaly (shortening of the cranium) and Scafocephaly (an elongation of the cranium) without Craniosynostosis being the cause of the deformations [5], [6].In the Netherlands the prevalence of SD was 13% at birth in 2001 with a decrease to 9.9% in 6 months after birth [7].

Skull Deformation

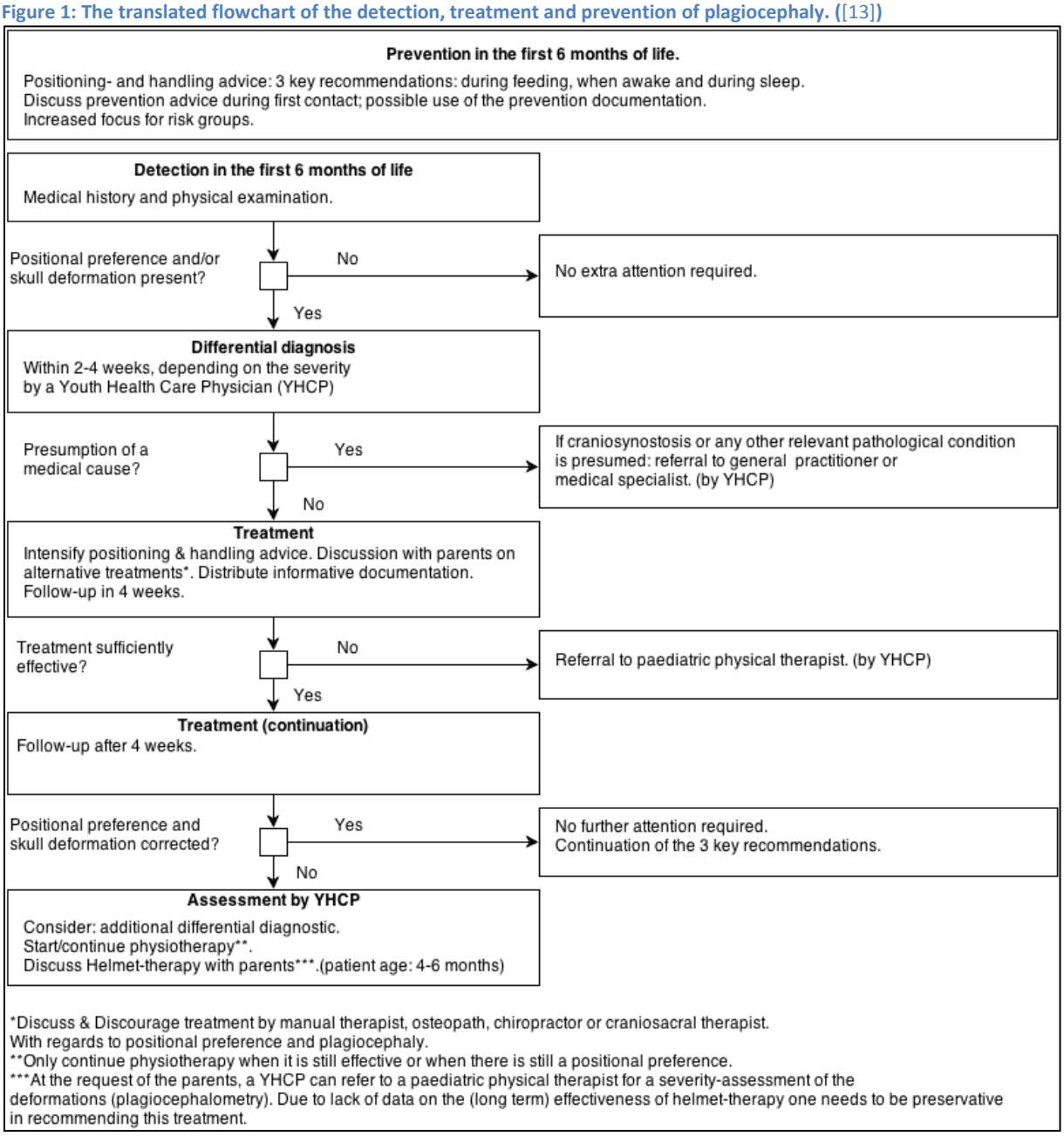
Skull Deformation at birth occurs commonly in human infants due to intra-uterine constrains and the natural malleability of the cranium. This natural malleability is a biological adaptation to ensure the relatively large human head can pass through the birth canal [8]. The gradual ossification of the bony plates and fusion of the membranous tissue (fontanelle) in between the plates are part of normal infant development and take up to 24 months to complete [9]. This accounts for the decrease in cranial malleability over time and the reason that any attempt to correct the head after one year becomes difficult. The deformations can also be worsened by external risk factors like vacuum –assisted delivery or the use of uterine forceps [10]. After birth children with a positional preference during sleep tend to either develop a skull deformation because their motor skill development is slow or the already present deformation is not corrected because of the positional preference [6].

Positional Plagiocephaly

The prevalence of positional plagiocephaly is hard to estimate as it changes with age. The point prevalence is documented to be peaking as high as 22.1% in the first 6 months of life.[11] Hutchinson *et al.* documented an overall drop in positional plagiocephaly at 2 years of age compared to neonatal cases.[6] Suggesting a normalization of the head shape in early childhood.[12] These numbers suggest that plagiocephaly is a common problem in new born infants that parents and physicians would have to deal with. In the Netherlands the deformation is typically noticed by the parents or the Youth Health Care Physician (YHCP) on the routine checkups. These medical professionals are part of the Dutch Infant Health Care Program which serves a monitoring and surveying role as opposed to the curative nature of health care [7]. In some cases the parents report their concerns to a general practitioner or directly to a paediatrician. If a deformation is present the YHCP examines the condition to exclude craniosynostosis, abnormalities in the sternocleidomastoid muscle, spinal growth

abnormalities and mobility related conditions. If the severity or cause of the plagiocephaly is related to one of these underlying conditions the patient is referred to a paediatrician for further examination and treatment. If the cause is a positional preference the patient is referred to a paediatric physical therapist. The parents are provided with information on the condition; and are instructed to use repositioning, 'tummy time' and environmental alterations to prevent worsening of the plagiocephaly. These measures are to prevent prolonged resting on the flattened area of the head and to balance the time spend on each side of the head [13]. In the first 3 months after birth the only treatment is trying to prevent a positional preference that would cause the deformations to occur or if already present worsen. From 2 till 6 months the addition of physical therapy can help to treat the deformation [14]. After 5 months the progress of the handling treatment the progress are observed by the treating physician. If no indication of improvement is observed, the physician can suggest the use of alternative treatments. Based on guidelines and personal expertise he/she can discuss the use of an orthotic helmet treatment. This helmet treatment would be used from 6 months and last until 12 months of age. When started after 8 months there is little chance of correcting deformations. This is why helmet treatment is not advised for new cases after this point [13]. See Figure 1.

Figure 1: The translated flowchart of the detection, treatment and prevention of plagiocephaly. ([13])



Severity Assessment

To assess the severity of the deformation, prominent experts in the field have tried to develop methods that could be used in defining the severity of the skull deformation using a general quantitative method. This has proven to be difficult as the skull deformation definition has inherent interpretive factors. The first choice assessment method was based on the method as described by Argenta (2004), where palpation and observation of the patient's deformations are compared with a predetermined template [15]. While this is the fastest and most non-invasive way of assessing the severity of the skull deformation it is also the most subjective method used. Another assessment method is Plagiocephalometry developed by van

Vlimmeren (2006). This method is used in the current assessment of skull deformation in the Netherlands [16]. This method uses a thermoplastic band to measure a variety of ratios related to the shape of the cranium. It is the only validated, easy-to-apply, method of diagnosing plagiocephaly. Other invasive and more expensive methods of assessment are for example x-ray scanning, MRI and Tomographic Imaging [17], [18]. These methods require a lot of time relative to plagiocephalometry and are therefore unpractical to use as a primary measuring method. This resulted in the recommendation to use Plagiocephalometry as a conclusive method when a skull deformation is suspected by an YHCP and rely on the Argenta Classification guidelines to detect skull deformations in the Netherlands.

Plagiocephaly Treatment

After the initial stages of the treatment there is a choice to be made as whether to continue positioning and handling or to start helmet treatment. This latter method has been criticised as a valid method of treatment on the basis that this method was never tested in a Randomized Controlled Trial (RCT) [19]. Methods that are not validated by this golden standard might still work their intended purpose but there is no supporting evidence that they will. Increasingly more medical policy makers require RCTs as evidence in their suggestions. These trials are used to research the effects of these medical devices, in this case the orthotic helmet used in plagiocephaly treatment.[20] The University of Twente started a RCT in 2009 called the HEADS (HElmet therapy Assessment in Deformed Skulls) Study. Ancillary to this study the methods and opinions of key actors in the treatment process were examined. These actors contained the parents, paediatric physical therapists and the helmet practitioners.

Study Goal and Research Question

To understand what Youth Health Care Physicians (preventive healthcare) and Paediatricians (curative healthcare) think about the current treatment of plagiocephaly and how they distinguish severity in skull deformations a questionnaire was used with clinical vignettes. These clinical vignettes will be the basis of a conjoint analysis where five attributes of severity and the physician's personal treatment preference will be researched.

The main research question is: "*Which characteristic of skull deformation is most influential in the perceived severity of the deformation; and to what extent does this perceived severity of skull deformation influence the preference for treatment of skull deformation in youth health care physicians and paediatricians in The Netherlands?*"

Methods

The five attributes are combined to display the severity of skull deformation in the clinical

vignettes. The two major plagiocephalometry indices: Oblique Diameter Difference Index (ODDI) and Cranio Proportional Index (CPI); two major visual deformations in the form of the Occipital Lift (OL) and the Ear Deviation (ED) and Gender. Some assumptions on the results can be made by looking more closely at these variables. The ODDI and CPI values are normally considered to be 100% and 80% respectively, so slight variations on these might not even be noticed. No amount of deformation on the ears and no occipital lift present are considered normal as the OL and ED values are more easily seen. Ear deviation cannot be corrected by the current helmet treatment as the helmet does not apply pressure on the temporal bones. Physicians aware of this limitation could not take into account the amount of ear deviation when considering helmet treatment.

It is plausible that males are at a higher risk of plagiocephaly than females [6], [11]. In general the male foetus is less flexible and the head is larger. In addition the male infant grows faster in the first 3 months [12]. Once a skull deformation is present the gender of the patient should not influence treatment.

Based on this information a hypothesis on the severity of skull deformation was formed.

"*The OL primarily and the ODDI and CPI secondarily are the three most influential attributes of skull deformation severity and the ED and Gender attributes are the least influential attributes.*"

Personal Treatment Preference is the preference a physician has for a specific treatment when more than one treatment is available. In the case of plagiocephaly the effects of the helmet treatment are not yet fully understood. Nor is it known what influences the physician's preference to a specific treatment. It is possible the severity of the plagiocephaly is influencing the personal preference to determine what the best treatment for each situation is.

This resulted in the following hypothesis for personal treatment preference: *"The treatment advice given for a patient with skull deformation is partially explained by the personal treatment preference of the physician but majorly influenced by the perceived severity of the skull deformation."*

Samplegroup

The target groups were Youth Health Care Physicians and Paediatricians. The (email) addresses were requested from the respective national professional organizations.

The Dutch Youth Health Care Physician Organization, the AJN (Artsen Jeugdgezondheid Nederland) provided the YHCP email address list which contained 705 e-mail addresses from YHCP that were working with children 0-4 years old. The Dutch Paediatrician Organization: NVK (Nederlandse Vereniging Kinderartsen) provided 1248 post addresses from paediatricians. Both unions reassured us these were from all Dutch health care professionals they had in their database.

From the Paediatrician list 35 entries were removed as their post address was not in the Netherlands, this selection process could not be used for the YHPC database containing the email-addresses.

Questionnaire construction

To investigate which of the attributes contribute the most to the perceived severity of skull deformation and to understand how this severity influences the treatment preference of the physician a questionnaire was constructed. The questionnaire was divided into two parts. The first part contained 62 general questions about the participant and their experience with skull deformation, handling and positioning treatment and helmet treatment. They were asked how they related to the parents and the concerns they might have and were asked to give their preference in treatment method for skull deformation. The second part was on the severity and treatment preference displayed in 10 clinical vignettes. Clinical vignettes have shown to be a valid way to test hypothetical scenarios in health care. [21] These vignettes contained 5 different attributes that were used in the conjoint analysis. The deformed head depictions are constructed from Argenta's classification graphical parts and plagiocephaly sheets gathered during the HEADS Study. Graphical editing software (Adobe Photoshop CS5) was used to put these parts and sheet together. The depictions of a deformed head contained the first 4 attributes: Oblique Diameter Difference Index (ODDI), Cranio Proportional Index (CPI), Occipital Lift (OL) and Ear Deviation (ED). A top-down representation was made as can be expected when using the outline of a plagiocephalometry band and a side view was drawn to display the OL. The Gender was described as text below the depictions which also contained the age of the 'patient' always being 5 months old. To understand how much this personal treatment preference influences the advice given two Likert scale questions were added to the questionnaire. The first scale question was to determine how severe the deformation in the depicted head was perceived in 5 steps from "None" to "Very Severe". The other question determined what kind of treatment the participant in this situation preferred. And this scale ranged from "Definitely Helmet" via "No Preference" to "Definitely Wait-and-See". Neither of these two scales had a way to opt-out of the question other then not filling in the question.

Conjoint analysis

The conjoined analysis was done on the 5 mentioned attributes which were randomized in PASW: SPSS 18 statistical software using an orthogonal design method. Via this method a total of 36 cases were generated which were divided over 4 versions. To each version a baseline scenario card was added depicting a "normal" head (100% ODDI and 80% CPI) with no deformities (No ED and No OL). This card was always the last card presented and was

added to check between versions for abnormal responses.

Attributes

The attributes for Severity were combined from four ODDI values and four CPI values. The ODDI values were 100, 104, 108 and 112; the CPI values were 80, 86, 92 and 98. To find plagiocephalometry cases matching these values a range of values were accepted for each level. To keep the amount of case variations to a minimum in order to prevent fatigue in the participant the OL, ED and Gender attribute each had 2 levels. Table 1 shows the exact values.

Table 1: The attributes and levels used in the conjoint analysis for the Severity. To maintain the properties of the plagiocephalometry bands used the ODDI and CPI values were approximated in a search range. This resulted in a head circumferences that came from actual plagiocephaly patients.

Attribute	Descriptive	Levels (Orders)			
ODDI	Intended Value (Searched Range)	100 (100-102)	104 (104-106)	108 (108-110)	112 (112-114)
CPI	Intended Value (Searched Range)	80 (80-83)	86 (86-89)	92 (92-95)	98 (98-101)
OL	Label (Value)	Yes (1)		No (0)	
ED	Label (Value)	Yes (1)		No (0)	
Gender	Label (Value)	Male (1)		Female (0)	

Table 2 shows the 16 found ODDI/CPI combinations used to depict the severity. The OL, ED and Gender levels were added to each combination of ODDI and CPI for a total of 128 combinations but reduced to 36 representative vignettes with the Principle of Orthogonal Design (POOD). The values were rounded down in the data analysis.

Table 2: The acquired ODDI/CPI combinations from the HEADS study database of plagiocephalometry bands

		ODDI			
		100	*104*	*108*	*112*
CPI	*80*	102,3-81,6	104,0-80,5	109,7-81,3	112,3-80,1
	86	100,0-88,9	106,3-86,3	108,1-86,7	112,7-88,6
	92	101,4-92,2	105,0-92,0	109,4-92,0	113,5-92,6
	98	100,8-98,5	105,5-98,4	108,7-98,3	113,0-98,0

Questionnaire Distribution

Before distribution the two part questionnaire was pilot-tested on two physicians and their feedback was used to improve the questionnaire. The questionnaire was distributed to the YHCP group via an online survey platform called SurveyMonkey. The paediatrician group got a paper copy of the full questionnaire. Both groups were informed

they could have the opposite method if they preferred, so the YHCPs could receive a paper copy and the paediatricians were given a link to their version on surveymonkey.com. After retrieval of the paper questionnaires the results were added to the responses on SurveyMonkey. Once the 3 month response period was over the data was downloaded from SurveyMonkey for further analysis. Participants who did not live in the Netherlands were excluded. Any incomplete surveys were discarded as none had completed the clinical vignettes which were a requirement for the study. Additionally the YHCP group had to be active practitioners with patients between 0-4 years old. The paediatrician group had to be active practitioners with plagiocephaly patients between 0-2 years old. The questionnaires were in Dutch.

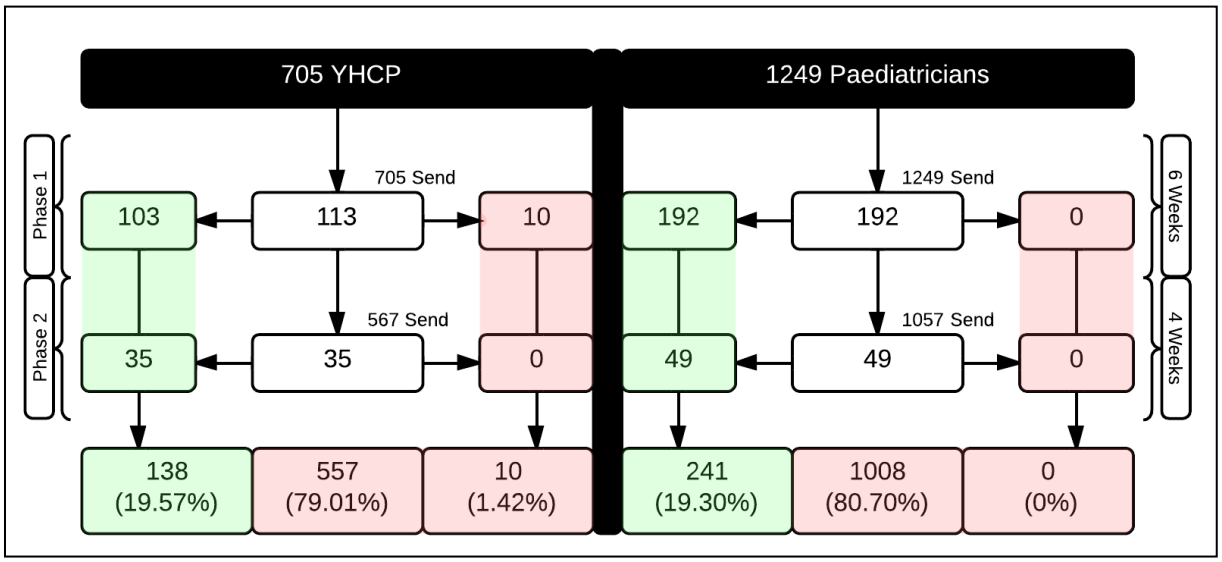
Data Analysis and Statistical Analysis

The data from the questionnaires was divided in General descriptive information about the participants, the perceived Severity of the depicted skull deformation by a participant and Treatment Preference the participant deemed appropriate in that situation. The Severity variable is a 5 point scale which consisted of the 5 attributes previously described. These attributes are considered to influence the severity and to understand what the largest influencing factor for treatment preference is a multivariable regression of the regressor variables X_1=ODDI, X_2=CPI, X_3=OL, X_4=ED and X_5=Gender with respect to the dependent variable (y)=Severity. The effects of Severity and (Unbiased) General Preference and Treatment Preference (per case) were checked with a correlation. The General Preference is the preference of the participant without a patient (case) to influence this preference. The Treatment Preference is the preference of treatment a participant after viewing a patient (or clinical vignette in case of this study) and is presented as a 5 point scale but the response was trichotomized into: Preference for Helmet Treatment, No preference, Preference for Wait-and-See treatment. The data was analyzed with the statistical software package IBM:SPSS Statistics v20.0.

Results

The questionnaire responses were gathered in two phases with a waiting period between them. See figure 2.

Figure 2: The data acquisition process in a diagram. Phase 1 and 2 were separated by a phase were the non-respondents were selected for a reminder. The values in white are the amount of responses, green represents the amount that actually was used in the study while red represents respondents that were excluded and whom did not respond.



The first phase lasted for 6 weeks and 113 YHCP responded (103 returned the questionnaire and 10 declined participation). A reminder was send to all 567 YHCP that did not respond. The second phase two lasted for 4 weeks. This yielded another 35 YHCP that returned the questionnaire (0 declined). After this no more questionnaires were returned or included. Resulting in a total of 138 (19.57% response rate) of YHCP. 20 YHCP respondents were not an active practitioner and were removed from the dataset after the initial descriptive analysis. Detailed distribution can be seen in table 3.

For the paediatricions a similar approach was taken. In phase one 192 paediatricians replied to the initial request with 0 declines. After sending out 1008 reminders another 49 (0 declines) responded bringing the total response rate on the paediatricians to 241 (19.30%). Of these 241 respondents 52 were not active practitioners or active on the topic of plagiocephaly. These were removed from the dataset after the initial descriptive analysis. (See table 3).

Table 3: The general descriptive analysis of the YHCP and Paediatrician respondent groups.

		Youth Health Care Physician		Paediatrician	
		N = 138	*%*	***N = 241***	*%*
Gender	Male	**3**	*2. 2*	**94**	*39.0*
	Female	**135**	*97.8*	**147**	*61.0*
Age	20-29 Years	**5**	*3.6*	**1**	*0.4*
	30-39 Years	**25**	*18.1*	**51**	*21.2*
	40-49 Years	**37**	*26.8*	**70**	*29.0*
	50-59 Years	**51**	*37.0*	**94**	*39.0*
	>/= 60 Years	**20**	*14.5*	**25**	*10.4*

Years of Practice	Missing	**20**	*14.5*		
	<5 Years	**14**	*11,9*	**34**	*18.0*
	5-14 Years	**27**	*22,9*	**72**	*38.1*
	15-24 Years	**47**	*39,8*	**40**	*21.2*
	25-34 Years	**29**	*24,6*	**41**	*21.7*
	>/= 35 Years	**1**	*0,8*	**2**	*1.1*
Experience with 0-4/0-2 year old patients	Yes	**135**	*97.8*	**190**	*78.8*
	No	**3**	*2.2*	**51**	*21.2*
Experience with "Wait and See" Treatment	Missing	**20**	*14.5*	**52**	*21.6*
	Yes	**113**	*95,8*	**176**	*93.1*
	No	**5**	*4,2*	**13**	*6.9*
Experience with "redressing helmet" Treatment	Missing	**20**	*14.5*	**52**	*21.6*
	Yes	**118**	*100.0*	**182**	*96.3*
	No	**0**	*0.0*	**7**	*3.7*
Advice Given based on personal treatment preference	Missing	**20**	*14.5*	**52**	*21.6*
	Neutral	**33**	*23.6*	**47**	*19.6*
	Yes	**62**	*45.1*	**129**	*53.6*
	No	**23**	*16.8*	**13**	*5.3*

Multivariate Regression and correlation

The constructed model for the perceived severity consisted of the ODDI, CPI, OL, ED and Gender variables. To find out which of these 5 have the biggest influence on the severity a multivariate regression analysis (MVR) was done where the Severity values were 0 (none) to 5 (very severe). The individual effects of each attribute can be seen in table 4.

Table 4: The beta values of each attribute in respect to the Perceived Severity. For both target groups.

$X_1 \dots X_5$	YHCP Beta	p	Paediatrician Beta	p
ODDI	0.367	<0.05	0.461	<0.05
CPI	0.212	<0.05	0.134	<0.05
OL	-0.246	<0.05	-0.324	<0.05
ED	-0.067	0.01	-0.036	0.135
Gender	-0.017	0.402	-0.130	0.588

The results of this MVR show that the biggest modifying value for Severity is the ODDI with the largest significant beta value for both respondent groups. The model used was also tested for the goodness-of-fit and this resulted in a R^2_a=0.392 for the YHCP and an R^2_a=0.298 for the Paediatricians. This means that the model of ODDI, CPI, OL, ED and Gender fits the Severity data of the YHCP group for 39.2% and 29.8% for the Paediatricians. The p-value for the attribute Gender was not significant modifier in both respondent groups. However the p-value for ED in the YHCP group was

below the significance value while it was not for the Paediatricians.

Table 5: The response to the clinical vignettes and the distribution of these responses ordered by each Attribute

Attribute	Level	YHCP Percentage	Paediatrician Percentage
ODDI	100	37,17	35,98
	104	19,49	19,83
	108	21,01	20,62
	112	22,32	23,57
CPI	80	33,04	34,77
	86	23,99	23,24
	92	20,22	20,12
	98	22,75	21,87
OL	Yes	53,33	54,19
	No	46,67	45,81
ED	Yes	54,78	53,44
	No	45,22	46,56
Gender	Male	50,00	50,00
	Female	50,00	50,00
Severity	1 (No Abnormalties)	9,78	10,62
	2 (Minor Abnormalties)	27,17	32,16
	3 (Moderate Abnormalties)	27,54	23,03
	4 (Major Abnormalties)	12,39	8,09
	5 (Severe Abnormalties)	1,67	0,79
	Missing	21,45	25,31
Preference	1 (Definitely Wait-and-See)	32,97	42,07
	2 (Probably Wait-and-See)	17,10	13,86
	3 (No Preference)	11,45	6,93
	4 (Probably Helmet)	12,39	7,97
	5 (Definitely Helmet)	4,64	3,86
	Missing	21,45	25,31

Respondent distribution

Table 5 shows the distribution of the attributes in the model. The distribution of respondents shows that a majority is more likely to wait-and-see when the perceived severity is low. See table for the YHCP. See figure 4 and 5.

Figure 3: The distribution of Treatment Preference compared to Severity for YHCP.

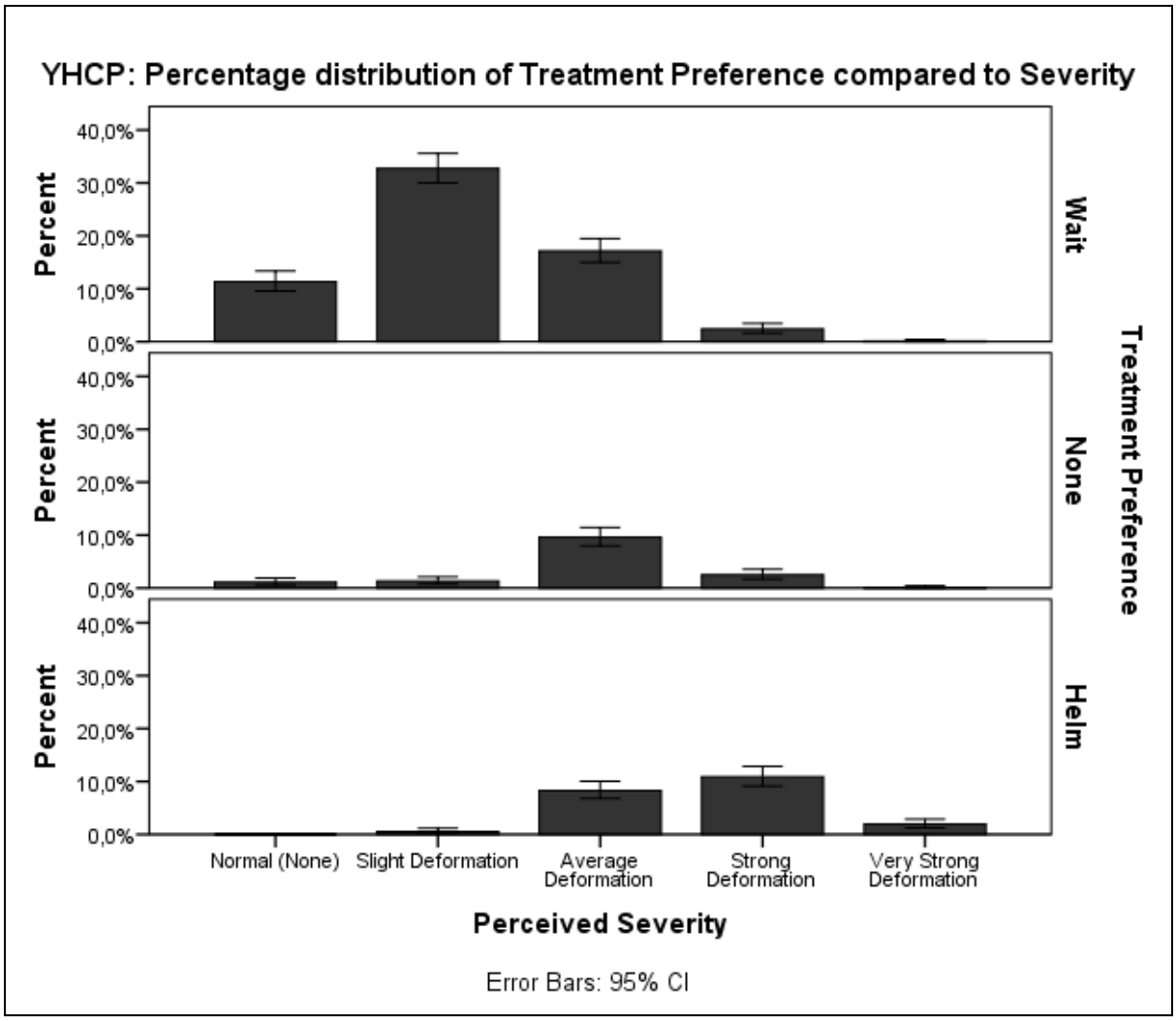
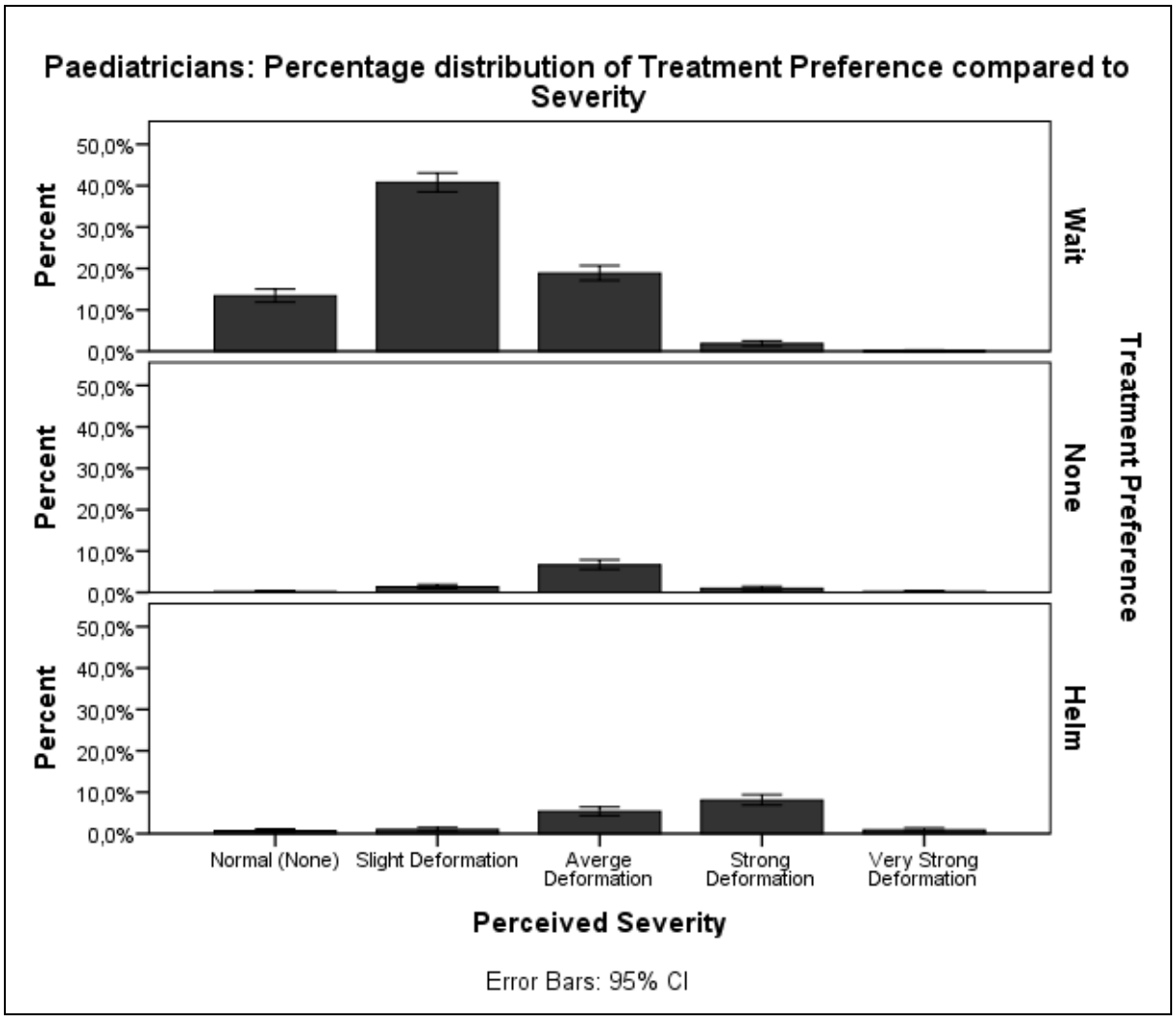


Figure 4: The distribution of Treatment Preference compared to Severity for Paediatricians.



Using severity as a single variable gives the option to see what the relation with treatment preference is via a correlation analysis. For the YHCP this correlation between Perceived Severity and Treatment Prefernce is R=0.717 (P<0.05) and for the paediatricians this correlation is R=0.633 (P<0.05).

Expanding on the idea that the General Preference is modified by the Severity to come to the Treatment Preference, a correlation between the general preference and the treatment preference was explored. This yielded R=0.243 (P<0.05) for the YHCPs and R=0.434 (P<0.05) for the paediatricians. The combined Severity and General Preference model where X_1 is Severity and X_2 is General Preference yielded a goodness-of-fit of R^2_a=0.572 for the YHCP and R^2_a=0.505 for the Paediatricians. Meaning that the model for both groups is explains about half of the dependent variable.

Discussion

The aim of this study was to understand how Youth Health Care Physicians and

Paediatricians view the treatment options of skull deformation. Which characteristics of skull deformation influence the perceived severity and to what extend does this perceived severity influence the treatment preference of the physician.

General Questionnaire

The results from the general questionnaire show that from both the participant groups the majority of the respondents know what a wait-and-see treatment entails. It also shows that this is the case for a helmet-treatment. This is indicating that both treatments are well established in both the first line (preventive) health care and the second line (curative) health care. The majority of the respondents were also giving treatment advice based on their own general treatment preferences. How this general treatment preference is formed is unclear and it was not tested by this questionnaire. However the assumption is that this is based on what information the physician is given about the treatment. The lack of RCT evidence on the precise effects of the helmet on plagiocephaly patients might explain the gravitation to the wait-and-see treatment as these values were overall significantly higher in cases where the severity was relatively low. Another factor to consider is the experience the physician has with both treatments, the amount of years experience with a specific treatment was not covered by the questionnaire however the majority of respondents did have at least 5 to even 25 years of experience in the field. It is therefore likely that the majority of respondents were active practitioners when the Dutch equivalent to the Back to Sleep campaign was introduced in the Netherlands.

Perceived Severity

From the results on the Perceived Severity it can be seen that the ODDI is the primary modifying factor and the occipital lift is the second largest factor. However the ODDI and CPI are interlinked with each other meaning that when the ODDI increases the CPI is likely to increase as well. It is not certain why the OL resulted in a higher contribution to the severity instead of CPI. This might be because of an error in the model or it could be that the “roundness” of the cranium is not considered as severe. Some studies suggest that asymmetry is considered less attractive in human-to-human interaction.[22] The hypothesis for Severity was based on the notion that occipital lift is a more striking deformation. The Ear Deviation and Gender attributes as these were the lowest contributing factors in both respondent groups. There are several reasons why the ear deviation could be the lowest contributor. For example the physician might know that the helmet-treatment does not work on the bones the ears are connected to. However it could also be that the shown deviations were not noted, meaning that the difference between the pictures with and without ED was not visible enough. According to previous research

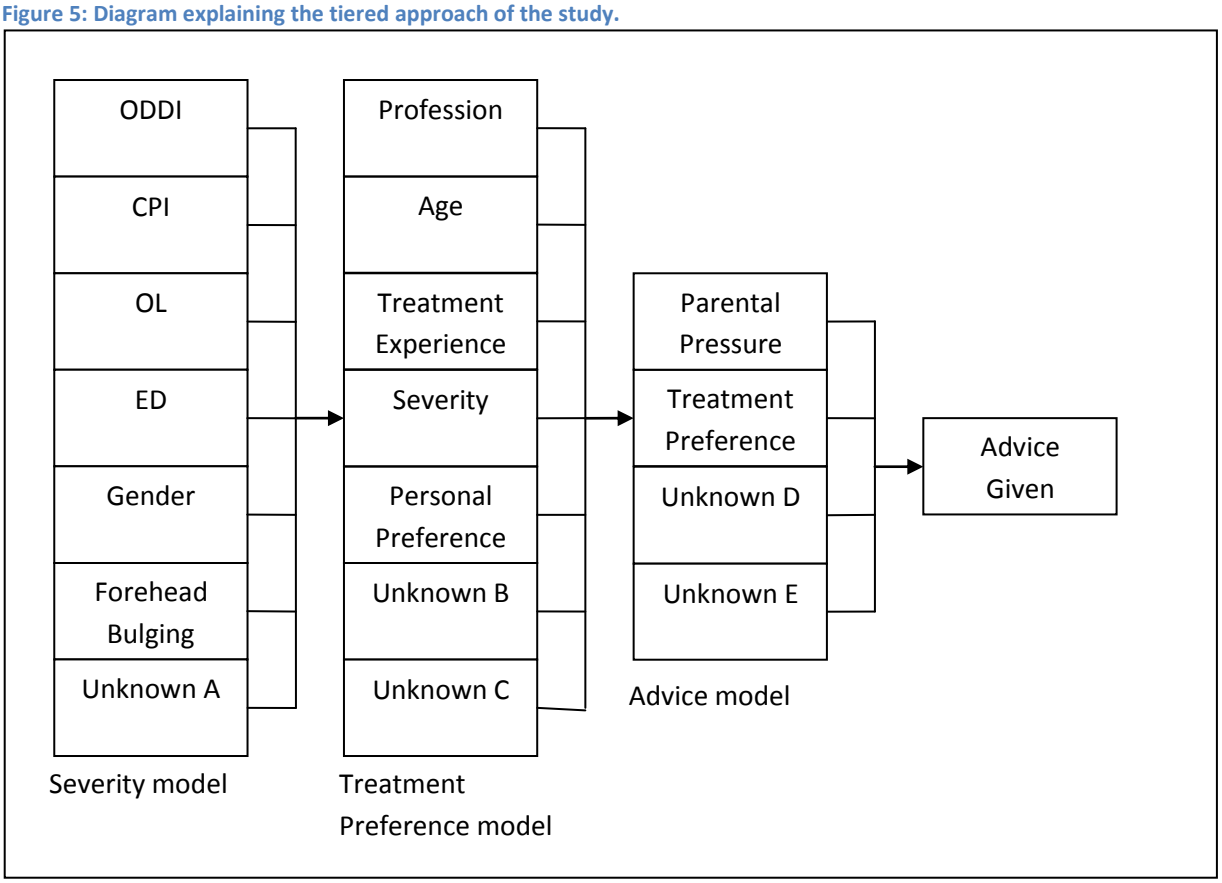
the male fetus is more likely to be born with skull deformations or acquire them during the first few weeks of growth. This however was not reflected in the perceived severity, as there was no significant difference in the severity between the male or female patients with skull deformation. There could also be considerations about the presentation of this attribute and this is discussed in greater detail in the clinical vignette section.

Treatment Preference

The result on treatment preference is showing a distribution that the higher the severity is rated the more likely the participants choose a helmet as possible treatment advice. This regression on this model is consistent with the distribution of the sample, so the assumption that Severity has a major role in treatment preference could be true. The personal treatment preference was also tested and while still being a positive influence on the treatment preference in the clinical vignettes its effects were less strong than the severity. As with the severity, the treatment preference is likely to be influenced by several additional variables besides the severity of the plagiocephaly and the physicians own personal preference. The majority of respondents seem to prefer the wait-and-see method but almost all respondents used a helmet when the deformations became more severe. The amount of paediatricians that preferred to use a helmet as treatment was lower than the YHCPs. The scope of this clinical vignette study was unable to test the influence of every possible variable. It is likely that some variables influence the General Preference, like the ODDI and CPI influence the Severity and that General Preference, Severity and other variables influence the Treatment Preference and subsequently the advice. In time a more fitting model could be extracted from testing additional variables to the ones done in this study. Other such variables could be hair length, motor-skill development speed. As can be seen in Figure 6, the Severity is considered to be combined from different variables related to the severity of plagiocephaly. The ODDI, PCI and ED are all part of the Plagiocephalometry method and the OL is also mentioned in the Argenta descriptions. Forehead bulging for example is a factor that was not considered in this study but is a part of the Argenta categorization method. Likewise there are indications that Severity alone is not the only factor to influence the model for advice. The personal preference as examined is such an additional factor. Also the age of the participant, the time spend with patients with plagiocephaly and the experience with the possible treatments are also likely factors that make up the treatment preference, or maybe the experience is part of the personal preference as the time spend familiarizing a treatment could bias the participant in preferring one treatment over another. And before the participant voices his advice to the parents there could also be external factors like the

voiced opinion or preference of the parents or the work methodology of colleagues.

Figure 5: Diagram explaining the tiered approach of the study.



The effect of both 'treatments' (wait-and-see and helmet-treatment) is essentially the same, the correction of a plagiocephaly within the time that the natural malleability of the cranium allows modification. If the rate of correction is the same in both treatments, it would not matter which treatment was used but the amount of deformation would be the limiting factor in the corrective effectiveness of both treatments. Based on the notion that wait-and-see is a very passive approach to correcting the skull deformation. The reduction of the positional preference would cause the skull deformation to decline in time. In that case the use of a helmet is not warranted from an economic viewpoint, as early detection of a positional preference could remove the plagiocephaly. It is of the utmost importance that these medical devices are tested on their effectiveness, preferably before a policy with financial implications is made. The results whoever show that higher severity tends to have a larger disposition to get treated with a helmet then a lower severity patient. This could indicate that the physicians are convinced that helmet treatment is either faster in correcting a certain amount of deformation per unit of time or that the helmet has the ability to correct a larger amount of deformation

compared to 'wait-and-see'. Whichever the case, once an RCT study is conclusive on the effects of the helmet treatment this distribution might chance.

Considerations on Clinical Vignettes

Compared to our clinical vignettes other studies that used Clinical Vignettes in general have a more broad set up with more attributes and less levels. This is done to create a more general, and arguably a more complete, representation of the described scenario.[21], [23], [24], [25] This option was not used in our case as the attributes were used from the plagiocephalometry classification method. The individual parts of the questionnaire and clinical vignettes that were used in this study could also have an effect on the results. For example in the clinical vignettes the gender was not integrated into the plagiocephaly picture but instead was written as an annotation below the pictures. It is not unlikely that this caused some respondents to skip over this information and not consider it when forming their opinion on the severity. A more prominent placement of the gender in either text or in picture form might have influenced the results. Also the clinical vignettes were in grayscale, the use of color might have alleviated this problem without changing the layout of the vignettes.

Another point to consider is the fact that the Clinical Vignettes were placed at the end of the questionnaire; this might have influenced the results as the respondents could suffer from the effects of fatigue.

Improvements on the models

The combinations of attributes that comprised the severity were also limited in options because the amount of questions had to be kept to a minimum. In the case of OL this resulted in a dichotomy instead of a range of values which an occipital lift can assume. The same goes for the Ear Deviation which in plagiocephalometry is a value in the millimeter range. For this research these nuances were not considered relevant to the goal but it is unknown what effect the actual measurements might have on the consideration process of the participant. The reasons for the clinical vignettes to have the severity depicted as a drawn figure based on the drawings of Argenta might also have an influence. The goal of these drawings was to make the severity as lifelike as possible; however the drawings are stylized representations of real patients. It is possible that a written text with information could trigger the participant's imagination and actually produce a better representation in their mind. It can also be that this happened with the stylized drawings as well. There is no way to control this effect other than using photographs of plagiocephaly patients with the appropriate values. Another consideration as to why the results might not have been comparable between YCHP and Paediatricians is because the approach to both respondent

groups was different. The paper variant of the questionnaire was distributed in an envelope that could contain an unfolded A4 sized stack of 28 pages. While the pilot testers could complete the full questionnaire within 30 minutes the size of the stack could have put many busy physicians off from taking the time to fill out the questionnaire. The same information was given to the YHCP group but they had a digital version which might look less daunting. On the other hand a digital distribution platform like SurveyMonkey might not work on all machines or in all work situations.

Conclusions

In conclusion the ODDI and OL values were the highest contributing factors of the set of variables we tested for in perceived severity. The hypothesis that predicted that the ear deviation (ED) and the gender were to be the lowest contributors was correct. The Treatment Preference of the physician is majorly influenced by the Severity of the plagiocephaly and less influenced by the preference the physician might have before seeing the patient. A likely reason for this is that the personal preference is the starting point of the decision process and is at the base of the choice the physician is making. It is unclear from this study if the treatment preference is always the same as the advice given. It is likely that this internal endpoint is influenced by external factors, like parental pressure, before the advice is given. To test this it might be an idea to actively influence the physician in a test environment by a parent or colleague. Another suggestion is to expand the amount of clinical vignettes, increase the OL and ED levels, and possibly remove the gender attribute to get a better indication of the effects of a multi-level Occipital Lift and Ear Deviation on Severity. Using actual photos of deformed heads and have a professional modify the pictures so they would exactly represent the underlying plagiocephalometry values would leave less room for interpretation from the respondent. Alternatively by using computer drawings a 3D image could be rendered where the effects of all variables could be individually generated and represented.

Acknowledgements

We would like to acknowledge the following persons. First R.M. van Wijk MSc for the use of her plagiocephalometry database. For the help with correcting SPSS syntax our thanks goes out to Dr. C.G.M. Groothuis-Oudshoorn. Also we acknowledge the help of Dr. J. Mourmans, Dr. F. Bunge and Dr. A. Meliesie with the pilot testing and contributing to the plagiocephalometry search. Lastly we want to thank everyone that helped in any way with this study but were not explicitly mentioned.

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