



Natural Progression of Non-Synostotic Plagiocephaly

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Thank you, today I will be presenting on my research on plagiocephaly.



Outline

- Background of Plagiocephaly
- Study Aims
- Methods
- Results
 - Head Shape Severity
 - Parent Satisfaction
- Future Research

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This presentation will go through a brief background of plagiocephaly, the reason for doing this study, how we did the study, and the results we got. I will then talk briefly about where the project is currently and where we plan to take it in the future.



What is Plagiocephaly?

- **Flattening** of the head ^{1(Littlefield 1998)}
- Types
 - Synostotic – due to premature closure of the skull sutures
 - **Non-synostotic** – due to external molding forces on the skull
 - Deformational plagiocephaly (DP)
 - Positional plagiocephaly



Plagiocephaly is a condition in which an infant's **head is deformed** as a result of prenatal and/or postnatal molding forces on the malleable and growing cranium. This results in asymmetrical flattening of the skull that has the shape of a parallelogram.

Plagiocephaly can be divided into two main categories: synostotic and non-synostotic. This study will examine non-synostotic plagiocephaly. This type of plagiocephaly is due to external molding forces on the skull, and there are no anatomic or internal issues preventing the skull from growing.

This picture shows a top view of a head with significant flattening on the right posterior of the skull, which is a typical presentation of plagiocephaly.

This is also called deformational plagiocephaly or positional plagiocephaly.



Incidence and Prevalence

- Prevalence of deformational plagiocephaly (van Vlimmeren 2007)
 - Varies with age
 - Birth – 13%
 - 6 weeks – 16%
 - 4 months – 19.7%
- Incidence has increased in the last 15 years (van Vlimmeren 2007)

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The prevalence of plagiocephaly has been shown to vary with age. More importantly, it has been shown that there has been a large increase in the number of cases of plagiocephaly being seen in the last 15 years.

This increase in plagiocephaly has been attributed to many factors. First and foremost the institution of the back to sleep program in 1992 by the American Academy of Pediatrics in an effort to decrease the incidence of SIDS. In addition to this, there has been the increased use of baby seats and interchangeable car seats, leaving kids on their backs for more time during the day. It is also thought that there may just be an increased awareness of plagiocephaly by doctors and parents so they are referring children more than before.



Treatment of DP

- Cranial Orthosis

- **Effective treatment** for improving head shape

(di Ribaupierre 2007, Graham 2005, Lee 2006, Littlefield 2000, Losee 2007, Loveday 2001, Plank 2006, Pollack 1997)

- **Standard of care** in the United States



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Various studies have shown that a cranial remolding orthosis or helmet, such as the one shown here, is an effective treatment for deformational plagiocephaly.

The helmet is considered the standard of care for deformational plagiocephaly in the United States. For this reason and the ethical considerations, it is difficult to create a randomized controlled study in the United States, and so far no such study has been undertaken to assess the effects of untreated plagiocephaly.



Study Aim

- To determine the **natural progression** of non-synostotic plagiocephaly of children between the ages of 18 and 48 months.
 - Hypothesis:
Children with deformational plagiocephaly **will not experience a natural improvement** in head shape.
- To determine the **parent satisfaction** with the child's current head shape.

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Despite knowing that there has been a large increase in plagiocephaly, and having an idea of what causes it, as well as knowing there is an effective treatment, there is still not a good idea of when plagiocephaly needs to be treated, or if it needs to be treated at all. Therefore, the primary aim of this study was to determine the natural progression of non-synostotic plagiocephaly.

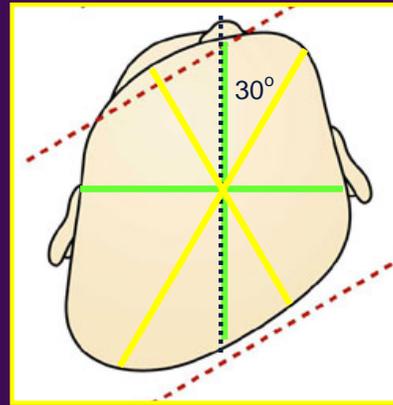
A second aim we had was to determine how satisfied parents were with the current shape of their child's head. This is important because even though the head may not change in shape as the child gets older, the growth of hair and the increase in the overall size of the head may make the flatness less obvious and therefore parents may find the head shape to be less of a concern later in life. It is also important because many parents struggle with the decision to treat their child for DP because of the many unknown variables that still exist.



Measurements

$$\text{Cephalic Ratio} = \frac{\text{Length}}{\text{Width}}$$

$$\text{Cranial Vault Asymmetry Index} = \frac{|\text{Diag A} - \text{Diag B}|}{\text{Larger (A or B)}}$$



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There are two main values that are used to diagnosis plagiocephaly.

The first is cephalic ratio which is measured as the ratio of the length to width.

The second is cranial vault asymmetry index. This is the difference between the length of two diagonals measured 30 degrees from midline, divided by the larger of the two diagonals. It is multiplied by 100 to create a percentage. This is commonly used because it normalizes the measurements allowing head shapes of various sizes to be compared. This was especially important for this study since the head size increased greatly during the years of growth.



Severity Scale

Severity Level	Description	CVAI	Suggested Treatment
1	<ul style="list-style-type: none">All asymmetry within normal limits	< 3.5	None
2	<ul style="list-style-type: none">Asymmetry in 1 posterior quadrant	3.5 – 6.25	Repositioning
3	<ul style="list-style-type: none">Asymmetry in 2 quadrantsMinimal ear shift, anterior involvement	6.25 – 8.75	Repositioning or Orthosis
4	<ul style="list-style-type: none">Asymmetry in 3 quadrantsModerate ear shiftAnterior involvement, orbital asymmetry	8.75 – 11.0	Orthosis
5	<ul style="list-style-type: none">Asymmetry in 4 quadrantsSevere ear shiftAnterior involvement, orbital and cheek asymmetry	> 11.0	Orthosis

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This severity scale was developed at Children’s Healthcare of Atlanta, and is based on data collected there. It has been shown that there is a correlation between the measured CVAI and the visual observations. Because orthotic treatment has not been recommended for levels less than a 3, we chose to include only those kids with a severity of 3 4 or 5 which would have been recommended for orthotic treatment. This created a subject population of children whose head shape was severe enough to require an orthosis but was not treated.



Methods – Subjects

- Twenty two subjects
- Inclusion criteria
 - Age between 18 and 48 months
 - Diagnosed with deformational plagiocephaly
 - Includes scan of head shape obtained before age 1 year
 - Declined treatment with cranial remolding orthosis
 - Severity of 3 or higher

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After doing a chart review of over 1000 charts, we identified 155 potential subjects. that are currently between the ages of 18 and 48 months. They had to be diagnosed with deformational plagiocephaly and had a scan taken before they turned 1 year old. They were included if they had a severity of 3 or higher and if they declined treatment with a cranial orthosis or helmet. They were each sent a letter followed by a phone call. Twenty seven parents agreed to have their child participate in the research project. One mother chose to only fill out the survey and not have the scan. 4 others were removed their initial scans were only severity 2. This left us with 22 subjects in this data set.



Methods – Survey

- Survey
 - Interventions
 - Parent impression

CHILDRENS HEALTHCARE OF ATLANTA
Patient Research Survey
Non-Synostotic Progression of Natural Plagiocephaly

Please answer the following questions about your child to the best of your ability.

Child's Name _____
Date of Birth _____ Current Age _____

Birth History
 Full term Premature, ____ Weeks
Complications (including time in NICU) _____

Health History
Has your child had any of the following?
 Torticollis, _____ with surgical correction
 Physical Therapy _____ for torticollis, ____ weeks
 for other _____
 Occupational Therapy
 Speech Therapy
 Hearing problems, please explain _____
 Frequent ear infections, how often? _____
 Ear tubes
 Vision problems, please explain _____
 Glasses
 Dental problems, please explain _____
 Surgeries, please explain _____

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During the visit, parents signed consent forms according to the approved IRB protocol. They then filled out a survey with questions regarding birth history, health history, family history as well as any interventions that they may have used such as repositioning and tummy time. Finally, there were two questions about how the parents felt the head shape had changed, whether it had improved, stayed the same, or worsened. And how satisfied they were with the current head shape, on a scale from very satisfied to not satisfied.

★★★ | Methods – Visual Observation



Digital photos were then taken of each subject.

First, a front, side and top view of the child as they presented that day with their hair. This gave us an idea of what the kids look like as they get older and have more hair to cover up any flatness that may be present.

We then placed an open-faced stockinette over their head took the same front, side and top pictures. This gives us an idea of what their head looks like without hair to cover it up.

These two pictures on the right show the top view, which is the easiest to see plagiocephaly. You can see the flatness much better in the bottom picture than you can in the picture with hair.

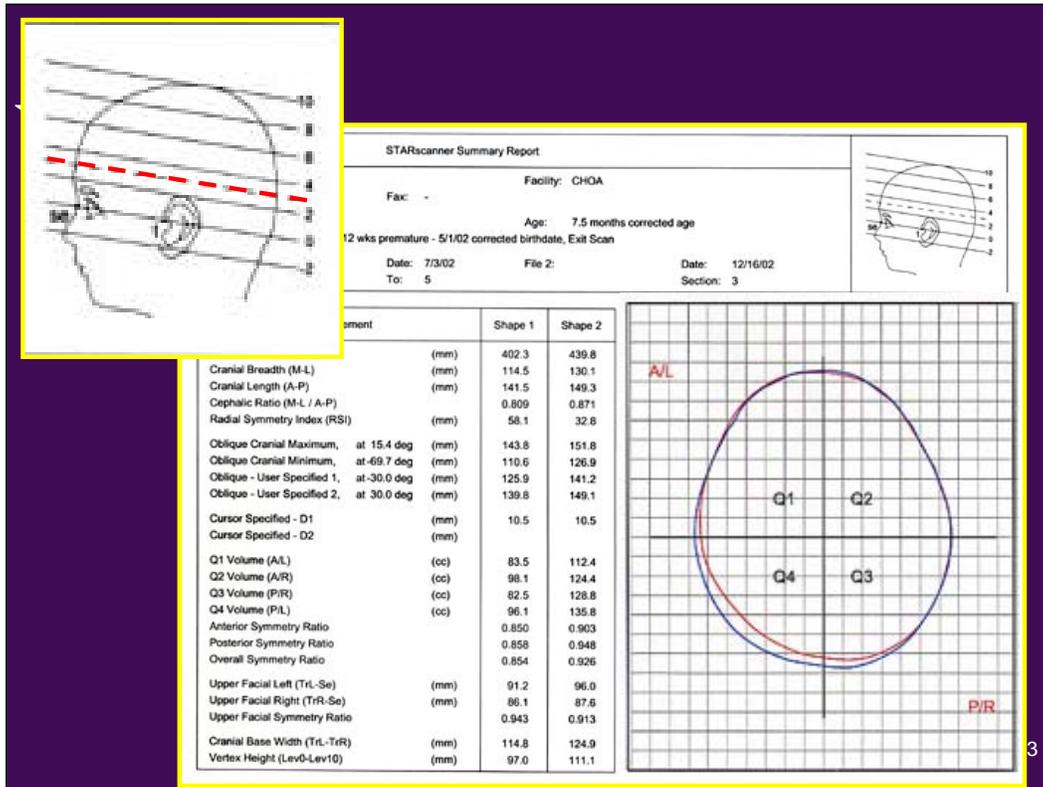


Methods – Scan

- **Re-scan** subjects using STARscanner
 - Compare variables with scan from time of initial diagnosis
 - Diagonal Difference
 - Cranial Vault Asymmetry Index
 - Cephalic Ratio
 - Severity Level

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We used the STARscanner laser data acquisition system to get measurements and calculations of the cranial vault asymmetry index and cephalic ratio. We also recorded the length of the diagonals measured at 30 degrees from midline and the severity level based on CVAI.



This is an example of a Starscanner summary report. The red line is the initial scan, the blue line is the follow-up scan. It is a cross section of the head just above the ears. It shows areas of growth, as well as reporting many measurements and calculations of head shape, including those used in this study.



Results – Head Shape

	Pre	Post	T-test ($p < 0.05$)
Diagonal Difference (mm)	12.9 ± 2.7	12.5 ± 3.4	$p = 0.58$
CVAI	8.4 ± 1.5	7.3 ± 1.8	$p = 0.02$
Cephalic Ratio	0.887 ± 0.05	0.844 ± 0.05	$p < 0.001$

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This table shows the mean values for the diagonal difference, CVAI, and cephalic ratio at the time of diagnosis (pre), and at the time of the study (post). What we've found is that there is no significant change in the difference of the length of the diagonals measured at 30 degrees. We attribute this to the fact that the sutures have closed, and so there should be growth every where around the skull, so if there is 6 mm of growth along one diagonal there should be an equivalent 6 mm of growth along the other diagonal. We did find a significant decrease in the CVAI of the subjects. However, it is noted that this decrease in CVAI was, in the majority of cases, not enough to move into a lower severity classification which will be discussed further in a moment.

One unexpected finding we had, was cephalic ratio. We found a significant decrease in the CR with an average decrease of 4%. What this means is that as kids are growing, their heads are getting longer more than they are getting wider. This is important clinically for children with brachycephaly, which is a flatness straight across the back of the head. Brachycephaly can occur on its own, or secondary to plagiocephaly. Our results suggest a natural decrease in cephalic ratio which could have an impact on current treatment protocols for these populations if examined further.



Results – Head Shape

		Post Severity Level			
		2	3	4	5
Pre Severity Level	3	30% n=3	50% n=5	20% n=2	0% n=0
	4	16.7% n=2	66.7% n=8	8.3% n=1	8.3% n=1

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This table shows the percentage of subjects who moved from one severity level to another. For example, 30% of subjects who were a severity 3 as infants, moved to a severity 2. The green boxes are those that improved in severity level, yellow stayed at the same level, and red worsened in severity.

This is useful clinically because it gives a way to tell parents who are in this level the chance of their child moving to another level. The usefulness of this table will increase as the subject pool increases.

What we've found is that 77% of all subjects improved in CVAI. However, only these 5 subjects here, representing 23% of the total improved enough to move them into a non-treatment severity level. So 77% of subjects whether they improved, stayed the same, or worsened in head shape would still be recommended for treatment with a cranial remolding orthosis.



Results – Parent Survey

- How do you feel your child's head shape has changed?

Improved	Stayed the same	Worsened	Don't Know
14	6	0	2

- How satisfied are you with your child's current head shape?

Very	Somewhat	Neutral	Not very	Not at all
11	6	0	5	0

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Our results from the survey found that the majority of parents feel the head shape has improved or stayed the same. Also, the majority of them are satisfied with the current head shape. And even the four parents who were not very satisfied with their child's current head shape, still reported that they felt it had improved.



Discussion – Quantitative

- Head shapes are improving in CVAI
 - 77% (17 of 22) of subjects CVAI improved
- However, **not improving enough** to move into a non-treatment classification
 - 77% of subjects would still be recommended for treatment
- Improvement in CR may indicate a less aggressive approach to brachycephaly treatment

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Overall, what we've found during this study is that head shapes are technically improving in their measured cranial vault asymmetry. We've seen that 77% of our subjects had an average decrease in CVAI of 1.6. However, this improvement is not enough to move them into a severity that would not be recommended for treatment. 77% of the subjects would still be recommended for treatment based on their current head shape. One unexpected finding from our data was that on cephalic ratio. We found that all subjects decreased in cephalic ratio an average of 4%. This is important clinically because if the brachycephalic head shape has some natural correction the minimum treatment level could be higher, however, this would need to be more extensively documented before any changes in treatment protocol were instituted.



Discussion – Qualitative

- **Parents are satisfied**
 - Hair
 - Growth
 - Justify decision
- Previous study (Steinbok 2007)
 - 77% of parents have no concern with the current shape of the child's head
 - 60% of parents feel the child's head shape is normal

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Further, we've found that parents are generally satisfied with their child's current head shape even with any residual asymmetry that may be present. They may be more satisfied due to the increased hair covering the flatness, growth that may make the flatness less apparent to them, or perhaps to justify their previous decision not to treat the plagiocephaly. It is also possible that their decision may be caused by a need to "please the doctor" so to speak, thinking that they are expected to be happy with the outcome.

A recent study found similar results to ours, reporting that 77% of parents have no concern with the current shape of their child's head, and 60% of the parents feel the shape is normal.



Future Work

- Expand sample size
 - Increase number of severe subjects
- Increase age range to look at additional time points
- Study other head shapes
- Multi-center study

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Based on our current results, we have a few future directions for the research. First, the sample size is continuously being increased. This study hasn't actually ended with 22 subjects. We would also like to see more ranges of severity. However, it is nearly impossible to find a severity 5 child who was not treated with the helmet. And ethically, it would be difficult to create a randomized study forcing some children who clearly need treatment into the non-treatment category. Second, we would like to increase the age range to look at further time points. We've seen a small improvement in the CVAI during the first few years of life, and we would like to see if this improvement continues.

Also, it has been proposed at Children's Hospital to continually scan children's heads at 6 or 12 month intervals after the time of diagnosis to assess changes. Since right now we can not guarantee whether the change in CVAI occurred before the fusion of the skulls plates, which is assumed to be before 18months, or if it is continually changing. We would also like to see the beginning of a multi-center study across the US. Finally, we would like to assess other head shapes including brachycephaly due to the finding in this study on cephalic ratio.

★ ★ ★ | Thank You!!!

Aaron Smith, CO





References

- American Academy of Pediatrics, Task Force on Sudden Infant Death Syndrome. The changing concept of sudden infant death syndrome: diagnostic coding shifts, controversies regarding the sleeping environment, and new variables to consider in reducing risk. *Pediatrics*. 2005;116:1245-1255.
- Di Ribaupierre S, Vernet O, Rilliet B, Cavin B, Kalina D, Lyevez PF. Posterior positional plagiocephaly treated by cranial remodelling orthosis. *Swiss Med Wkly*. 2007;137:368-372.
- Graham JM, Gomez M, Halberg A, et al. Management of deformational plagiocephaly: repositioning versus orthotic therapy. *J Pediatr*. 2005;146:258-262.
- Lee WT, Richards K, Redhead J, Papay FA. A pneumatic orthotic cranial molding helmet for correcting positional plagiocephaly. *J Craniofac Surg*. 2006;17(1):139-144.
- Lima D. The management of deformational plagiocephaly: a review of the literature. *J Prosthet Orthot*. 2004;16(4S):9-14.
- Littlefield TR, Beals SP, Manwaring KH. Treatment of craniofacial asymmetry with dynamic orthotic cranioplasty. *J Craniofac Surg*. 1998;9:11-17
- Littlefield TR, Pomatto JK, Kelly KM. Dynamic orthotic cranioplasty: treatment of the older infant. Report of four cases. *Neurosurg Focus*. 2000;9:1-4.
- Littlefield TR. Car seats, infant carriers, and swings: their role in deformational plagiocephaly. *J Prosthet and Orthot*. 2003;15(3):102.



References

- Losee JE, Mason AC, Dudas J, Hua LB, Mooney MP. Nonsynostotic occipital plagiocephaly: factors impacting onset, treatment, and outcomes. *Plast Reconstr Surg.* 2007;119:1866-1873.
- Loveday BPT, de Chalain TB. Active counterpositioning or orthotic device to treat positional plagiocephaly? *J Craniofac Surg.* 2001;12(4):308-313.
- Moss SD. Nonsurgical, nonorthotic treatment of occipital plagiocephaly: what is the natural history of the misshapen neonatal head? *J Neurosurg.* 1997;87:667-670.
- Plank LH, Giavedoni B, Lombardo JR, Geil MD, Reisner A. Comparison of infant head shape changes in deformational plagiocephaly following treatment with a cranial remolding orthosis using a noninvasive laser shape digitizer. *J Craniofac Surg.* 2006;17(6):1084-1091.
- Pollack IF, Losken HW, Fasick P. Diagnosis and management of posterior plagiocephaly. *Pediatrics.* 1997;99:180-185.
- Rekate H. Occipital plagiocephaly: a critical review of the literature. *J Neurosurg.* 1998;89(1):24-30.
- Steinbok P, Lam D, Ginsh S, et al. Long-term outcome of infants with positional occipital plagiocephaly. *Childs Nerv Syst.* 2007;23:1275-1283.
- van Vlimmeren LA, van der Graaf Y, Boere-Boonekamp MM, L'Hoir MP, Helden PJ, Engelbert RH. Risk factors for deformational plagiocephaly at birth and at 7 weeks of age: a prospective cohort study. *Pediatrics.* 2007;119:408-418.



Questions



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Thank you for listening, and are there any questions?



Severity Scale

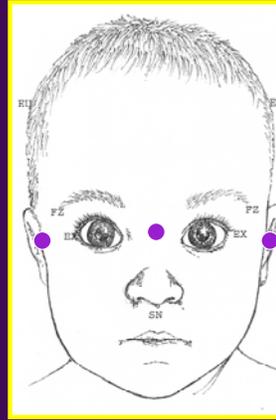
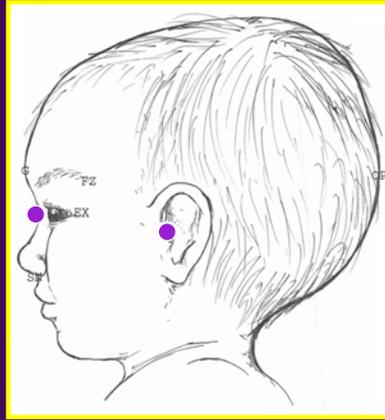
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Markers for measurements





Effects of Growth on CVAI



Diagonal 1 = 136.5 mm
Diagonal 2 = 149.3 mm
Difference = 12.9 mm

$$\text{CVAI} = \frac{12.9}{149.3} \times 100 = 8.64$$



Diagonal 1 = 175.2 mm
Diagonal 2 = 188.1 mm
Difference = 12.9 mm

$$\text{CVAI} = \frac{12.9}{188.1} \times 100 = 6.85$$

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★★★ | Brachycephaly





Why the increase in DP?

- **Back to Sleep Program** (AAP 1992)
- Use of baby swings, bouncy seats, **interchangeable** car seats and child carriers (Littlefield 2003)
- Decrease in use of soft bedding (Littlefield 2003)
- Decreased time in prone position (Littlefield2003)
- **Increased awareness** of DP by parents and caregivers (Rekate 2007)

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This increase in plagiocephaly has been attributed to many factors. First and foremost the institution of the back to sleep program in 1992 by the American Academy of Pediatrics in an effort to decrease the incidence of SIDS. In addition to this, there has been the increased use of baby seats and interchangeable car seats, leaving kids on their backs for more time during the day. It is also thought that there may just be an increased awareness of plagiocephaly by doctors and parents so they are referring children more than before.



Who cares?

- Children
- Parents
- Orthotists
- Third party payers

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While we now have an idea of what causes plagiocephaly, and we know that the helmet treatment works to correct the deformity, the question remains who cares what the head can do on its own? First, the children because they will need to be in the helmet and it is their permanent head for the future. Second, parents because they need to make the final decision of whether to undergo treatment or not. It is of concern to orthotists because with the huge increase in the number of kids they're seeing with plagiocephaly, perhaps they are treating kids who don't need to be treated or vice versa. Finally, payers are interested because, well frankly they are the ones paying for the treatment. So they want to know that it is needed.



Results – Parent Survey

- **Torticollis**
 - 11 out of 22
 - Physical therapy
 - 11 out of 11

	Torticollis	No Torticollis
Improved	10	7
Worsened	1	4

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Another finding we had from the survey was that 50% of the subjects had been diagnosed with torticollis which is known to have an effect on plagiocephaly. All children who were diagnosed with torticollis also received physical therapy to treat it. What we found was that 10 out of these 11 children actually improved in CVAI. This could likely be connected to the treatment of the torticollis.