Treatment of unilateral coronal synostosis by endoscopic strip craniectomy or fronto-orbital advancement: Ophthalmologic findings

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BACKGROUND Unilateral coronal synostosis results in ipsilateral retrusion of the forehead and superior orbital rim, shortening the elevation of the orbital roof and contralateral frontal bossing and orbital roof depression. This craniosynostosis is associated with the development of V-pattern strabismus and aniso-astigmatism. Since 2004 endoscopic strip craniectomy performed in patients by 3 months of age has been offered as an alternative to fronto-orbital advancement at 9 to 11 months of age. We compare the incidence and severity of V-pattern strabismus and aniso-astigmatism in children treated by these 2 procedures.

METHODS A retrospective review identified 37 children with unilateral coronal synostosis treated with either fronto-orbital advancement or endoscopic strip craniectomy. Incidence and severity of V-pattern strabismus, fundus excyclotorsion, and aniso-astigmatism was recorded for an "early" examination (between 2 and 14 months of age) and a "late" examination (between 14 and 45 months of age).

RESULTS Early examination revealed no statistical difference in severity of V-pattern strabismus or aniso-astigmatism between the 2 groups. At late examination there was a trend toward greater severity of V-pattern strabismus, an increase in excyclotorsion, and a statistically significant increase in the standard deviation of aniso-astigmatism in the cohort of children treated by fronto-orbital advancement.

CONCLUSIONS Children with unilateral coronal synostosis treated by early endoscopic strip craniectomy may develop less severe V-pattern strabismus, excyclotorsion, and range of aniso-astigmatism than those treated by later fronto-orbital advancement. This is an early, retrospective, nonrandomized study with a short follow-up period; longer follow-up is necessary to confirm these results. (J AAPOS 2009;13:155-160)

Unilateral coronal synostosis, the second most common single suture synostosis, is frequently associated with the development of V-pattern strabismus. The synostosis restricts and redirects orbital growth ipsilaterally, resulting in compensatory frontal bossing and overgrowth contralaterally. The associated strabismus and ocular torticollis have been attributed to these orbital changes.[1-8] The majority of patients also develop aniso-astigmatism and, accordingly, may have an increased risk of amblyopia.[4,9] Although correction of unilateral coronal synostosis by fronto-orbital advancement (FOA) improves the orbital symmetry anterior to the axis of the globe, some posterior asymmetry may persist. Furthermore, this procedure often is deferred in patients until later infancy (9 to 11 months) to reduce anesthetic risks and the likelihood of recurrent frontal asymmetry.[10] Perhaps for this reason, established dysmotility and aniso-astigmatism do not typically resolve.

Recently, an older technique, strip craniectomy,[11] has been revitalized with an endoscopic approach.[12] Typically performed by 3 months of age, endoscopic strip craniectomy (ESC) removes the fused coronal suture under endoscopic guidance; a helmet is used to direct brain growth to gradually improve cranial symmetry.

Since 2004, patients at Children's Hospital Boston have been treated with both techniques. This has provided an opportunity to assess whether an earlier and different operation might reduce the severity of V-pattern strabismus and aniso-astigmatism in patients with unilateral coronal synostosis. We present a retrospective study comparing the incidence and severity of V-pattern strabismus and aniso-astigmatism in children with unilateral coronal

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synostosis treated by endoscopic strip craniectomy and helmeting with those treated by fronto-orbital advancement.

Materials and Methods

Ophthalmic records of children referred to the Department of Ophthalmology between 2002 and 2007 with a diagnosis of unilateral coronal synostosis were reviewed. The following clinical features were confirmed: ipsilateral retrusion of the forehead and superior orbital rim, shortening and elevation of the orbital roof, nasal root angulation and anterior displacement of the ear, and malar eminence. Contralateral findings included frontal bossing and depression of the orbital roof. All patients had confirmatory computed tomography (CT), demonstrating fusion of the involved coronal suture with possible extension to the fronto-sphenoid suture (Figure 1). Excluded were patients with CT evidence of fusion of other cranial sutures, those not examined in our department at or before 14 months of age, or those who chose to have surgical correction and follow-up at other centers.

Patients referred for craniofacial evaluation before 4 months of age had been given the option of choosing either ESC and helmeting or FOA. Parents were told that ESC was a less complex procedure but carried the risk of anesthesia at a younger age and possible recurrence of synostosis (reossification) and need for later FOA. Infants referred after 4 months of age underwent FOA, a more invasive cranial reconstructive procedure, typically performed between 9 and 11 months of age. The choice of procedure depended only on infant age and parental choice and not on severity at presentation or any other clinical features.

All ophthalmic records of eligible patients were reviewed and data on the sensorimotor evaluation, fundus torsion (when documented), and cycloplegic refraction were collated. On the basis of these data, an ophthalmological summary of V-pattern strabismus, fundus torsion, and aniso-astigmatism was derived for each patient for an "early" examination and a follow-up, or "late," examination.

For patients treated with ESC, the "early" examination was based on data from ophthalmological evaluation before 12 months of age and within a range of 2 to 11 months. For the FOA group, the "early" examination occurred in the range of 3 to 14 months of age. The "late" ophthalmological examination was completed after the patient's first birthday and always after FOA or ESC. It took place for the ESC group between 14 and 45 months of age and for the FOA group between 12 and 37 months. One patient from each group was not included in the analysis of the "early" examination because he or she was not seen at or before 14 months of age. All patients were examined at times appropriate for the "late" examination and therefore included in primary outcome analysis.

Strabismus was graded in the following manner. Full motility examination was reviewed, and versions were graded for the amount of overelevation and underdepression in adduction. This strabismus was classified as mild (0 to 1/2+), moderate (1 to 2+), and severe (3 to 4+) V pattern.[13] Fundus (anatomical) torsion was graded based on the appearance of the posterior pole examined with indirect ophthalmoscopy and a 20 D lens in conditions of dim illumination, while the infant's head was untilted and when gazing at a visual stimulus at the far end of the examination room.[14] When these conditions were met, the degree of torsion was determined by the ophthalmologist at the time of the examination. E-Supplement 1 (available at jaapos.org), provides a sample of fundus photos illustrating the appearance typical for patients with varying degrees of anatomical torsion. These photos, however, were not used as a "standard" in this retrospective study.

Table 1 summarizes criteria for designating severity of V-pattern strabismus and fundus torsion. Cycloplegic refractions had been performed 30–45 minutes after instillation of 2 drops of 1% cyclopentolate hydrochloride by an ophthalmologist within our department. Refraction data were later used to calculate anisometropia and aniso-astigmatism. Aniso-astigmatism was calculated with the use of plus cylinder astigmatism; measured astigmatic error of the contralateral eye was subtracted from the astigmatic error measured in the ipsilateral eye.[8] The spherical anisometropia was calculated by subtracting the spherical equivalent of the contralateral eye from the spherical equivalent of the ipsilateral eye.

Statistical Methods

Separate analysis was performed for both the early and late visits to assess the relationship between severity of V pattern strabismus (mild, moderate, or severe) and type of craniofacial repair. Because there were very few examples of severe dysmotility at the early visit, moderate and severe categories were combined. A χ^2 test was used to test for an association between treatment group and the degree of V-pattern strabismus. For the late visit there were more children with a severe V pattern, so the moderate and severe categories did not need to be combined. An association between the treatment group and the degree of V-pattern strabismus was analyzed using the Cochrane-Armitage trend test.[15]

To assess aniso-astigmatism, the difference in the means was tested with the use of one-way analysis of variance, with severity of aniso-astigmatism as the response and treatment group (ESC or FOA) as the predictor. Age at visit was considered as a covariate but ultimately discarded, as it added no extra information to the model. To test for differences in the variances, point estimates were obtained between the 2 groups, *s2 FOA* and *s2 ESC*. The data were then "bootstrapped" to obtain 95% and 99% confidence intervals.[16] If these confidence intervals did not overlap, this was equivalent to rejecting a test that the variances of the groups are equal at a 5% (for the 95% CI) and 1% (for the 99% CI) significance level. Separate analysis was performed for the early and late visit.

Results

Forty-eight patients initially were identified with characteristic clinical and radiographic features of unilateral coronal synostosis. Of the 48 patients, 11 were eliminated on the basis of exclusion criteria noted previously: 5 had additional fused sutures; 4 did not meet our age requirements for ophthalmic evaluation; and 2 had surgical treatment elsewhere. Of the 37 patients included in the study, 11 underwent endoscopic strip craniectomy at a mean age of 3 months (range, 2-5 months) and 26 had a fronto-orbital

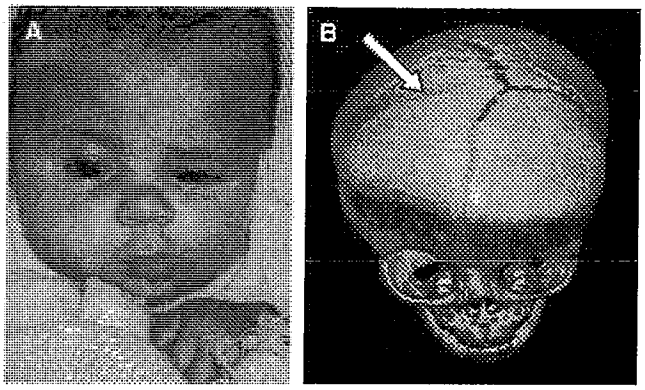

FIG. 1. (A) Classic features of unilateral coronal synostosis. Note ipsilateral elevation of the brow and retrusion of the roof of the orbit, contralateral frontal bossing and diminution of the interpalpebral fissure, ipsilateral twist of the nasal root, and contralateral point to the chin. (B) Arrow indicates the fused right coronal suture on reformatted CT imaging.

Table 1. Categorization of severity of V pattern strabismus and fundus (anatomical) torsion

Category	V-pattern strabismus	Fundus torsion
Mild	0 to 0.5+	Less than or equal to 1+
Moderate	1 to 2+	1.5 to 2+
Severe	3 to 4+	> 2+ to −4+

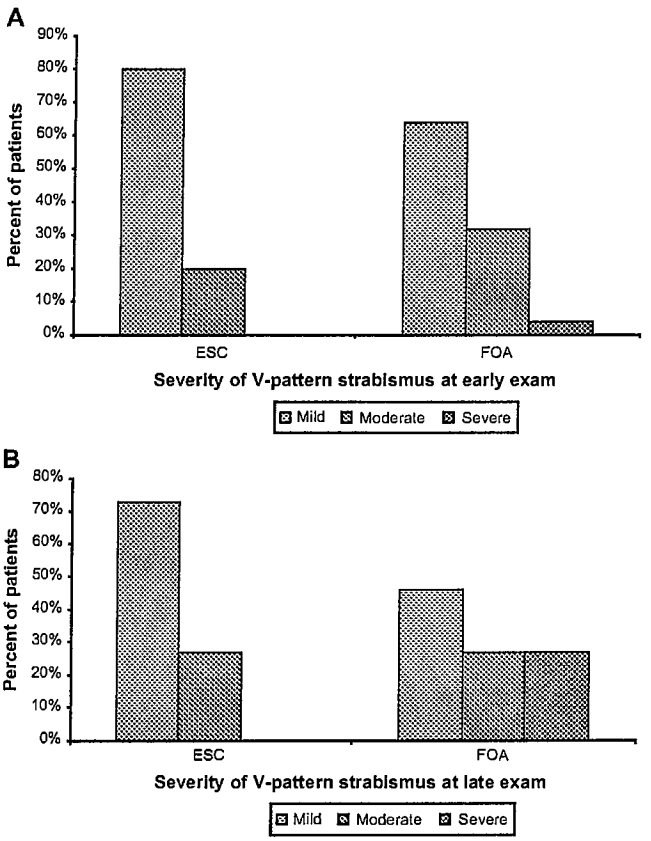

FIG. 2. Severity of V-pattern strabismus by type of primary craniofacial repair (ESC or FOA): severity at early exam (A); severity at late exam (B).

advancement at a mean age of 11 months (range, 8-18 months). The age range and mean age for the early examination of the patients undergoing FOA (3-14 months; mean, 7.56 months) was slightly later than that for the early evaluation of the ESC group (1.5-11 months; mean, 5.85 months); this was a natural outcome of the later referral age of many patients ultimately scheduled for FOA. The difference in age at evaluation was, however, not statistically significant. The follow-up or "late" ophthalmological examination took place on average, at 20.54 months of age for the ESC group, and 22.5 months of age for the FOA group. Neither the mean age nor the range of ages at which the late (and primary outcome) examination was performed was statistically different for our 2 study groups.

In e-Supplement 2 (available at jaapos.org), classification of severity of V-pattern strabismus is noted under "motility" and degree of anatomical torsion under "torsion" for early and late ophthalmic examination. Age and type of craniofacial repair, need for secondary correction, and any strabismus procedures performed are included in the clinical profile. Figure 2A summarizes the motility outcomes at the early ophthalmic examination visit; Figure 2B, at the late ophthalmic examination for the ESC and FOA groups.

The χ^2 test for an association between treatment group and severity of V-pattern strabismus at the early examination was not significant ($p > 0.05$), indicating that at this stage, the 2 groups were essentially the same in this parameter. The Cochrane-Armitage trend test for an association between treatment group and severity of motility at the late examination gave a p-value of 0.056. Seventy-three percent of those treated with ESC had no or mild V pattern, 37% had a moderate V pattern, and none had a severe V pattern. Of those treated with FOA, 46% had no or a mild V pattern, 27% had a moderate V pattern, and 27% had a severe V pattern. Although these findings were significant only to a p value of 0.056, the difference noted between the 2 groups is suggestive of an effect and warrants further study.

V-pattern strabismus often correlates with anatomical (fundus) excyclotorsion, so it was evaluated as well. Accurate information on fundus torsion was obtainable or recorded on very few patients at the early examination (10 of 11 patients undergoing ESC and 10 of 26 patients undergoing FOA). More data were available for the late ophthalmic examination (11 of 11 patients undergoing ESC and 15 of 26 patients undergoing FOA). Because the percentage of patients with recorded data on this outcome were too small for meaningful statistical analysis, we note only that the outcomes measured were consistent with the previous motility findings. A greater percentage of patients who were treated with FOA demonstrated moderate-to-severe fundus torsion than those

Table 2. Fundus excyclotorsion at early and late examination for unilateral coronal synostosis patients treated with endoscopic strip craniectomy and helmeting (ESC) or fronto-orbital advancement (FOA)

Primary craniofacial repair	Exam timing	Mild	Moderate	Severe
ESC	Early, n = 10	9/10	0/10	1/10
	Late, n = 11	9/11	2/11	0/11
FOA	Early, n = 10	4/10	1/10	5/10
	Late, n = 15	6/15	4/15	5/15

Table 3. Evolution of V-pattern strabismus at early and late examination for unilateral coronal synostosis patients treated with endoscopic strip craniectomy and helmeting (ESC) or fronto-orbital advancement (FOA)

Primary craniofacial repair	Exam timing	Mild	Moderate	Severe
ESC	Early, n = 10	9/10	0/10	1/10
	Late, n = 11	8/11	3/11	0/11
FOA	Early, n = 25	16/25	8/25	1/25
	Late, n = 26	12/26	7/26	7/26

who underwent ESC. Table 2 shows that 1 of 10 patients undergoing ESC and 6 of 10 patients undergoing FOA showed moderate-to-severe excyclotorsion at the early examination, and whereas 2 of 11 patients undergoing ESC and 9 of 15 patients undergoing FOA demonstrated moderate-to-severe excyclotorsion at the late examination.

Because we did note a trend toward greater V pattern and increased fundus excyclotorsion in patients treated with FOA (Table 3), we thought it would be informative to compare the frequency and type of surgical correction of strabismus for these 2 populations over an equivalent follow-up period. The median age of our population of patients treated by ESC at their last exam was 18.5 months, the mean was 24 months, and the range was 12 to 45 months. For patients treated by FOA, the median age was 50 months, with a mean age of 59 months, and a range of 12 to 147 months. The incidence of strabismus repair in our patients treated with ESC (2 of 11) compared with the incidence of strabismus repair performed in the FOA group by 21 months of age (5 of 26) or 30 months of age (8 of 26) is not statistically different. It is noteworthy, however, that all patients in the FOA group requiring strabismus repair had surgery for their V pattern; the only V pattern necessitating repair in the ESC group was for a child with reclosure of the coronal suture and need for subsequent FOA.

Refractions for the first and second ophthalmic visits as well as calculated aniso-astigmatism and aniso-sphere are detailed in e-Supplement 3 (available at jappos.org). As noted previously, 2 patients, one from each group, did not have early examinations.

Furthermore, 2 additional patients from the FOA group were not cooperative enough at their first evaluation for accurate refraction. There was no statistically significant difference in mean aniso-astigmatism and aniso-sphere between patients treated by FOA and those by ESC at either the early (Figure 3) or late ophthalmic evaluation. In both the ESC and FOA groups, mean aniso-astigmatism was negative, confirming the previously reported tendency toward greater astigmatism in the eye contralateral to the synostosis.[9,17] At the first examination, mean aniso-astigmatism was −0.5 for the ESC group and −0.38 for the FOA group. At the second examination, mean aniso-astigmatism was −0.28 for the ESC group and −0.41 for the FOA group. Nevertheless, there was a statistical difference in the standard deviation or the "spread" of aniso-astigmatism, i.e., a larger spread readily apparent in those treated by FOA. The magnitude of aniso-astigmatism (positive and negative) measured was likely to be greater in patients treated with FOA ($p < 0.01$), even though the mean was similar to those treated by ESC.

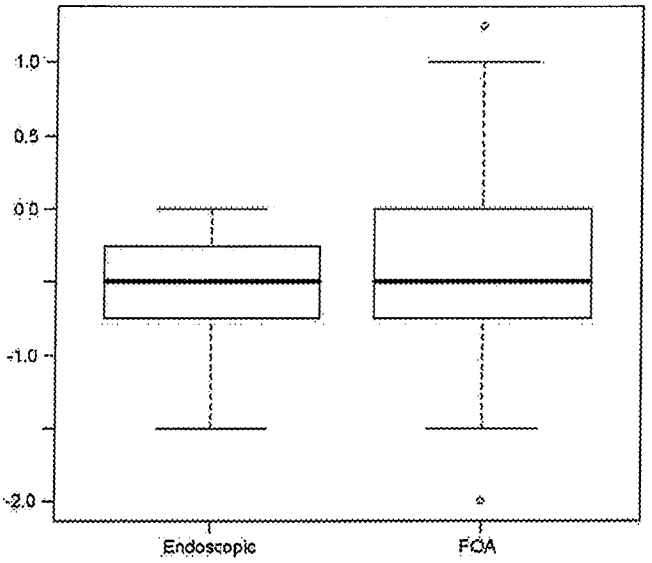

FIG 3. Aniso-astigmatism derived from refractive error measured at early examination in children with unilateral coronal synostosis treated with either front-orbital advancement (FOA) or endoscopic strip craniectomy (ESC).

The confidence intervals for the variances of the 2 groups (ESC, FOA) just abutted for the first examination (95% CI, 0.045-0.340 for ESC, and 0.337-0.900 for FOA) and did not overlap for the second examination (95% CI, 0.062-0.212 for ESC, and 0.420-2.42 for FOA). Thus the standard deviations are statistically different at the 0.05 percent level in the early examination, and the standard deviations are statistically different at the 0.01 percent level in the late examination. Larger amounts of aniso-astigmatism were more common in the group of children treated by FOA.

Discussion

In this retrospective review comparing ophthalmic outcomes of patients with unilateral coronal synostosis treated

by ESC and helmeting in early infancy to those corrected in late infancy by FOA, we report a trend toward decreased severity of V-pattern strabismus in those treated with early ESC and helmeting. We found there was a reduction in anatomical excyclotorsion, although the number of patients reliably evaluated for this finding was much smaller. Finally, although both groups exhibited significant aniso-astigmatism, the FOA group had a statistically greater standard deviation ($p < 0.01$) or spread in magnitude of aniso-astigmatism, as compared with those treated by ESC and helmeting.

This preliminary study has a number of limitations, including unrealized selection bias, a short follow-up period, an unequal number of patients in each treatment group, a relatively small number of patients, and some disparity in age at assessment. Whereas lesser severity might have unwittingly biased the craniofacial team toward performing ESC in those presenting early enough to be considered for either procedure, those presenting for evaluation later might, on the whole, have been a less severely affected cohort. The first potential bias would increase the number of "less severely affected" children in the ESC group and the second would increase the number of less severely affected children in the FOA group. With respect to selection bias, our craniofacial team is unaware of any difference in the severity of unilateral coronal synostosis that distinguishes the group of patients treated by ESC from those by FOA. According to these physicians, age at presentation and parental preference were the only determinants of primary intervention. More patients were treated with FOA because many presented too late to be considered for ESC, and some parents were unwilling to consider a "newer" procedure.

Because this is a retrospective study, some examinations (2 motility and refraction evaluations for the early examination period and 26 of 74 torsion assessments) were not performed or were unobtainable and not included in our analysis. The follow-up period has been relatively short, ranging from 14 to 45 months; disparity in age at assessment was attributed to the retrospective nature of this study. Most ophthalmic examinations were performed before conception and initiation of this retrospective chart review, and ophthalmic outcomes were unlikely to have been biased; the examiners were not, however, masked with respect to the type of craniosynostosis repair.

Estimates vary on the incidence of V-pattern strabismus, relative "superior oblique palsy" or hyper-elevation in adduction with unilateral coronal synostosis. In a retrospective study by Robb and Boger,[2] 27 of 33 unilateral coronal synostosis patients had this finding. Seven of 14 unilateral coronal synostosis patients evaluated prospectively in a study by Gosain and colleagues[8] had V-pattern strabismus, and 57.6% of 59 patients in a retrospective study by MacIntosh and colleagues[4] had this pattern. In our retrospective series combining those treated with ESC and those with FOA, we found that 22 of 37 (60%) had at least 1 + V-pattern strabismus and, of these, 10 had ipsilateral hyper-elevation in adduction, 11 had bilateral hyper-elevation in adduction, and 1 had contralateral hyper-elevation in adduction. One patient had hyper-elevation in abduction. We found that 6 patients had primary position esotropia, 2 had primary position exotropia, and 1 an A pattern.

Fronto-orbital advancement may place the patient at a motility disadvantage for two reasons. First, this procedure is performed months after dysmotility has begun to occur with resultant secondary changes in sarcomere length in the involved muscles.[18] An earlier repair with ESC may be advantageous. Second, fronto-orbital advancement displaces the bandeau anteriorly after stripping free the periorbita; in this process, the trochlea likely falls further behind, reinserting passively. This may exacerbate the relative retropositioning of the superior oblique in patients with unilateral coronal synostosis or, at the very least, do little to repair this problem. Inferior oblique function may become relatively stronger as its origin remains unchanged with this procedure. If this explanation of extraocular muscle imbalance is true, ESC with helmeting may allow for both earlier resolution of this asymmetry in function between superior oblique and inferior oblique, and may prevent a mild iatrogenic superior oblique palsy.

With respect to the effectiveness of FOA at resolving relative "superior oblique palsy," Diamond and colleagues[19] found that 11 of 34 patients developed new-onset V-pattern strabismus after FOA, which spontaneously resolved in only 1 of 34. Gosain and colleagues[8] noted spontaneous resolution of strabismus in only 1 of 8 with preexisting strabismus after FOA, the onset of new strabismus in 1 patient of 15 having FOA, and a need for surgical correction of strabismus in 7 of 9 after FOA. They do not discuss impact of FOA on the "severity" of preexisting strabismus.

With respect to timing of the procedure, Denis and colleagues[20] report a decrease in incidence of strabismus from 67% to 19% by moving up the mean age of corrective FOA from 23 months to 5.6 months. This finding suggests that timing may be more important than the technique used. Early FOA has been associated, however, with relapse of the orbital position and retrusion necessitating a need for secondary repair; it is not, therefore, favored by craniofacial surgeons.[21-23] The option of early ESC may be preferable.

In contrast to patients with unilateral coronal synostosis, resolution or reduction of V-pattern strabismus has been reported with FOA and correction of exorbitism in cases of Apert or Crouzon syndromes. It has been postulated that surgical resolution of retrusion of the inferior orbit and resolution of exorbitism reduces direct contact of the inferior oblique with the globe, and thus reduces inferior oblique overaction.[24,25] Because patients with unilateral coronal synostosis have neither exorbitism nor retrusion of the inferior orbit, there should not be enhanced contact of the inferior oblique with the floor of the orbit. Accordingly, FOA for unilateral coronal synostosis should have no direct effect on this proposed mechanism of V-pattern strabismus.

We report a significantly greater standard deviation in aniso-astigmatism in patients with unilateral coronal

synostosis undergoing FOA at 9 to 11 months of age as compared to those managed earlier by ESC. The result is greater aniso-astigmatism in the FOA group, and a higher risk of amblyopia. In both groups, the mean level of aniso-astigmatism confirms the conclusions of Levy and colleagues[9] and Tarczy-Hornoch and colleagues[17] that there is greater astigmatic error in the eye contralateral to the synostosis. The greater delay in orbital repair and subsequent increase in contralateral frontal bossing may be responsible for development of a greater degree of astigmatism in the FOA group.

This retrospective study compares the potential impact on the evolution of V-pattern strabismus, excyclotorsion, and astigmatism in patients with unilateral coronal synostosis treated by early ESC and helmeting to those treated by FOA. There is a trend suggesting that the former may help prevent severe V pattern and excyclotorsion, and there is a statistically significant reduction in severity of aniso-astigmatism. We caution that this is an early, retrospective study with a relatively short follow-up period. Evaluation of a larger number of patients with longer follow-up is needed to confirm or refute this difference in outcome.

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