



# Why do metopic sutural synostoses angulate? The concept of nasion sutural complex and its implication on the management of hypotelorism—early results and proof of concept

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## Abstract

**Object** Angulation at the suture is a hallmark of metopic synostoses amongst all craniosynostoses. No other sutural synostoses demonstrate angulation at synostoses consistently. We look into the possible aetiology and the implication of the understanding in the treatment goals of trigonocephaly. We hypothesise that the nasal bone and nasofrontal suture viz. “nasion sutural complex” are involved in trigonocephaly along with the well-accepted role of metopic suture. We propose that it is the angulation at this junction which leads to trigonocephaly and its secondary features.

**Materials and methods** The study included seven infants, who underwent correction for trigonocephaly at our paediatric craniofacial division at Amrita Institute of Medical Sciences and Research Centre, Kochi, India, between the period July 2015 to March 2018. The cohort included were infants with trigonocephaly who had CT head for diagnosis. We analysed the multidimensional CT (MDCT) of these infants and compared to an equal number of age-matched controls. The controls were infants with other forms of sutural synostosis with metopic uninvolved and normal infants where MDCT was done for other reasons. Sutural characteristic at the nasion and metopic suture recorded in comparison with an equal number of age-matched controls. We performed spring cranioplasty for three infants after metopic suturectomy, extending the release beyond the nasion sutural complex, placing springs to distract the suture. The infants who underwent spring cranioplasty were followed up for the aesthetic outcome. Remaining infants of the study underwent standard frontorbital correction for metopic craniosynostoses.

**Results** We could demonstrate a fusion of nasofrontal and nasal suture in all cases ( $n = 7$ ) of trigonocephaly included in the study on MDCT and intraoperatively. We performed spring cranioplasty for three infants ( $n = 3/7$ ), where we released the internasal suture. At 3 months follow-up, along with correction of the angulation, the hypotelorism improved significantly. Other infants in the study (4/7) underwent classical frontorbital advancement.

**Conclusions** Fusion of nasion sutural complex along with metopic sutures may explain the angulation in trigonocephaly. We propose that all minimally invasive techniques for correction of trigonocephaly and associated hypotelorism should consider this fact for an improved outcome.

**Keywords** Metopic suture · Craniosynostoses · Trigonocephaly · Spring cranioplasty

## Introduction

Angulation at the suture is a hallmark of metopic synostoses. *No other synostoses demonstrate consistent and predictable*

*angulation at the abnormal suture.* We look into the possible explanation and its implication on treatment goals.

## Materials and methods

The study included seven infants, who underwent correction for trigonocephaly at our paediatric craniofacial division at Amrita Institute of Medical Sciences and Research Centre, Kochi, India, between the period July 2015 to March 2018. These infants were identified from a cohort of 90 children operated during the same period for craniosynostoses.

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**Table 1** Details of the patient who underwent spring cranioplasty for trigonocephaly and outcome

S. no.	Age/sex	Intercanthal distance (pre-op/post-op) (22.22 + − 1.13)*	Lateral orbital distance (pre-op/post-op) (65.56 + 1.76)*	Interdacryon distance (pre-op/post-op) (14.16 + 0.74)*	Outcome
1.	3 m/F	2.30 cm/2.67 cm	5.8 cm/6.6 cm	1.3 cm/1.5 cm	Did not undergo the secondary procedure given the underlying cardiac anomaly. The spring was removed after 3 months. Follow-up of 20 months
2.	4 m/M	1.97 cm/2.07 cm	6.05 cm/6.5 cm	1.12 cm/1.19 cm	Underwent spring removal after 3 months with cranial vault remodelling. This infant had associated scaphocephaly. Follow-up 15 months
3.	3 m/M	2.13 cm/2.58 cm	6.24 cm/7.62 cm	1.19 cm/1.64 cm	Underwent spring removal at 6 months. Follow-up was 14 months

\*Normative value [2]

#### Inclusion criteria:

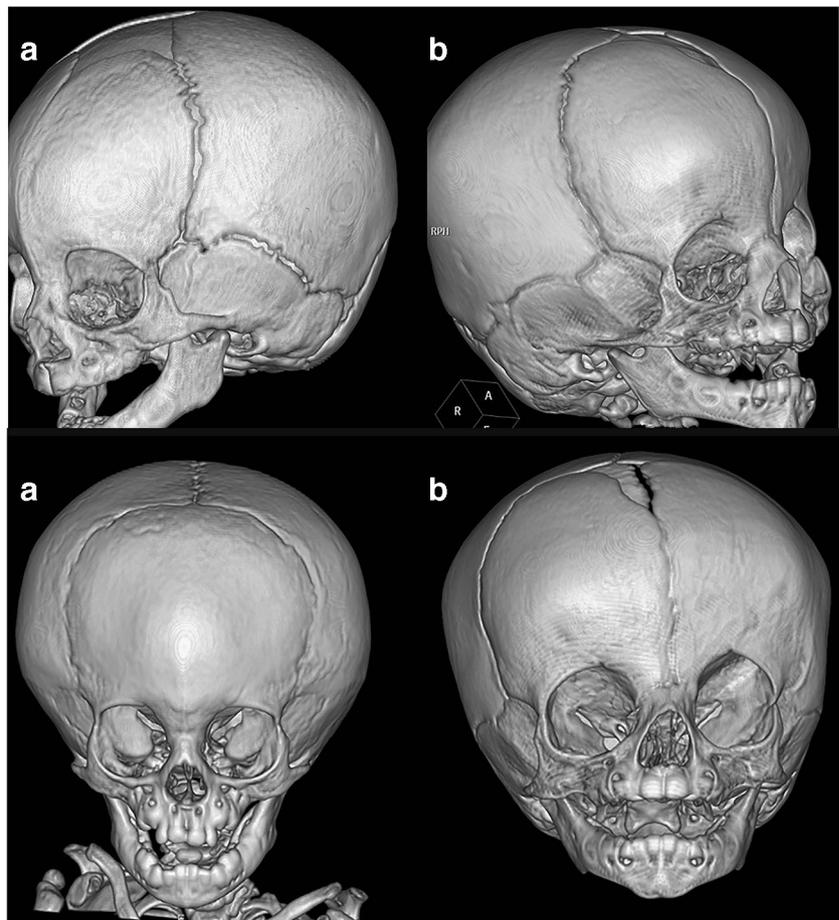
- 1) All infants with trigonocephaly with CT demonstrating metopic sutural synostoses ( $n = 7$ ). Routine CT is not a part of our treatment protocol for simple craniosynostoses. Hence, these infants had CT elsewhere and were referred or CT done with a diagnostic possibility of multisutural synostoses. We analysed the multidimensional CT (MDCT) of these infants and compared to an equal number of age-matched controls who were infants

with other forms of sutural synostosis without the involvement of metopic and normal infants where MDCT was done for some other indication.

Sutural characteristic at the nasion and metopic suture were recorded in comparison with equal number of age-matched controls.

- 2) Of these three infants ( $n = 3$ ) (average age of 3.3 months) underwent spring cranioplasty. The three infants chosen had definite trigonocephaly and hypotelorism compared to normative standards

**Fig. 1** (Upper panel) **a** MDCT of a three-month infant (infant 1) with trigonocephaly with fused metopic suture. Note the fused nasofrontal and nasal suture viz. “nasion sutural complex.” The CT also demonstrates the compensatory changes viz. hypotelorism, temporal hollowing. Compare with a control infant in the right with unicoronal synostoses **b** showing open metopic, nasofrontal, and nasal suture viz. “nasion sutural complex.” This infant (infant one ref. table) did not undergo further remodelling, but only spring removal at 3 months. (Lower panel) Lateral view of the infant (infant 1) in **a** with trigonocephaly **b** normal control infant with open “nasion sutural complex”



available [1, 2]. Other infants ( $n = 4/7$ ) underwent standard frontobital advancement.

We performed metopic suturectomy, and we extended the release beyond the nasion sutural complex in three ( $n = 3$ ) patients and placed springs to distract the suture (Table 1, details of the patient who underwent spring cranioplasty for trigonocephaly and outcome). The infants were followed for the aesthetic outcome. The average follow-up was for 18 months. The springs were removed at 3–6 months. The other four infants underwent routine frontorbital advancement. We do not use helmets during the postoperative follow-up.

### Results

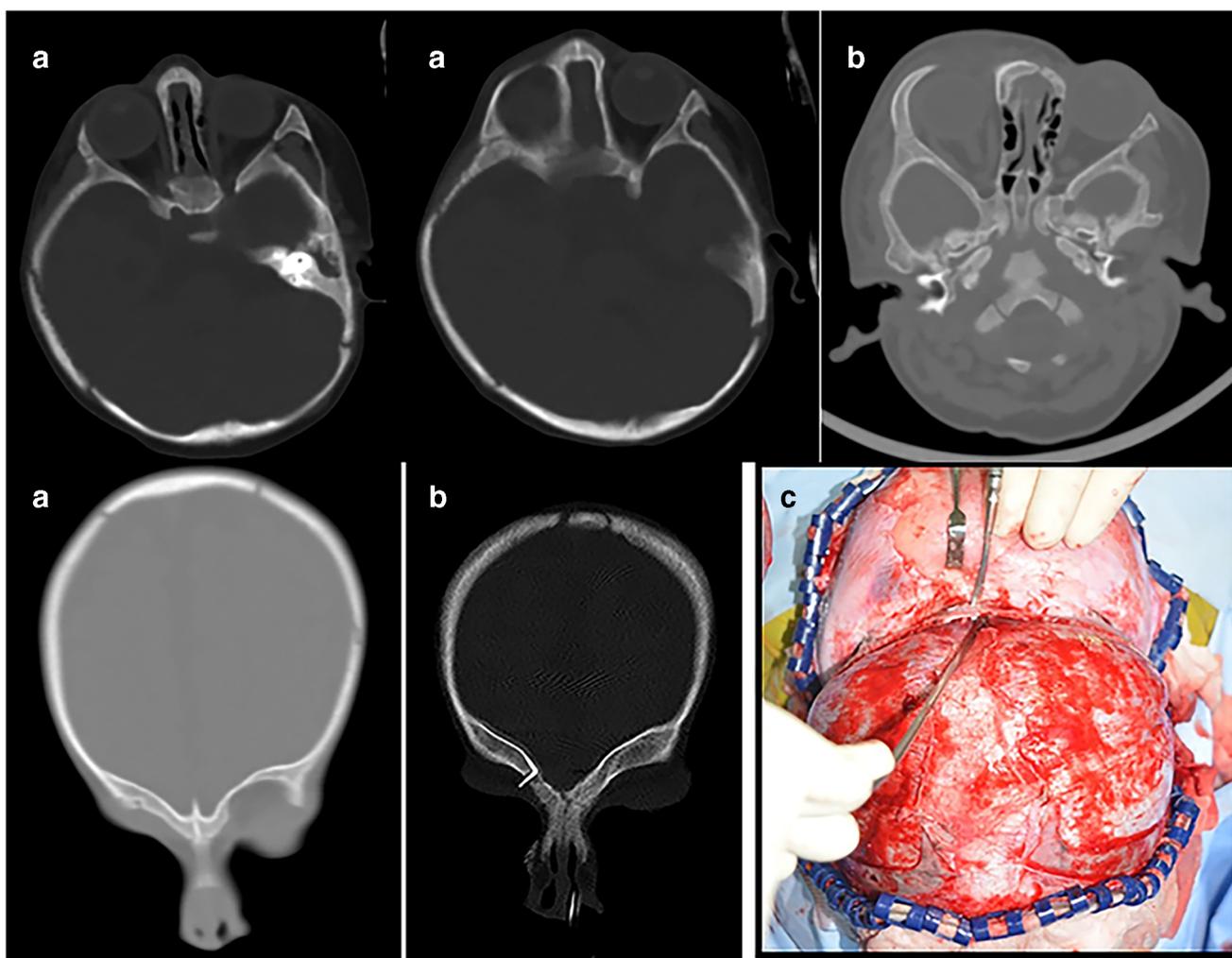
The study included seven infants with clinically confirmed trigonocephaly, and CT confirmed metopic sutural synostoses.

The age range was 3 to 12 months. We noticed fusion of nasal suture and nasofrontal suture in all cases of metopic synostoses with trigonocephaly (Fig. 1). The nasal suture involvement was observed radiologically and seen intraoperatively in all patients (Figs. 1 and 2).

We performed spring cranioplasty for three infants ( $n = 3/7$ ) with trigonocephaly. The distraction was performed with a customised spring with one of the springs placed just above the level of the nasion and the second higher in the released metopic suture. The average duration of surgery was about 90 min with an average blood loss of approx. 15 ml.

At 3 months of follow-up, along with the correction of the angulation, the hypotelorism improved significantly and remained stable (Figs. 3 and 4) (Table 1). The average follow-up was for 15 months (range 14–20 months). We do not use helmets during the follow-up period.

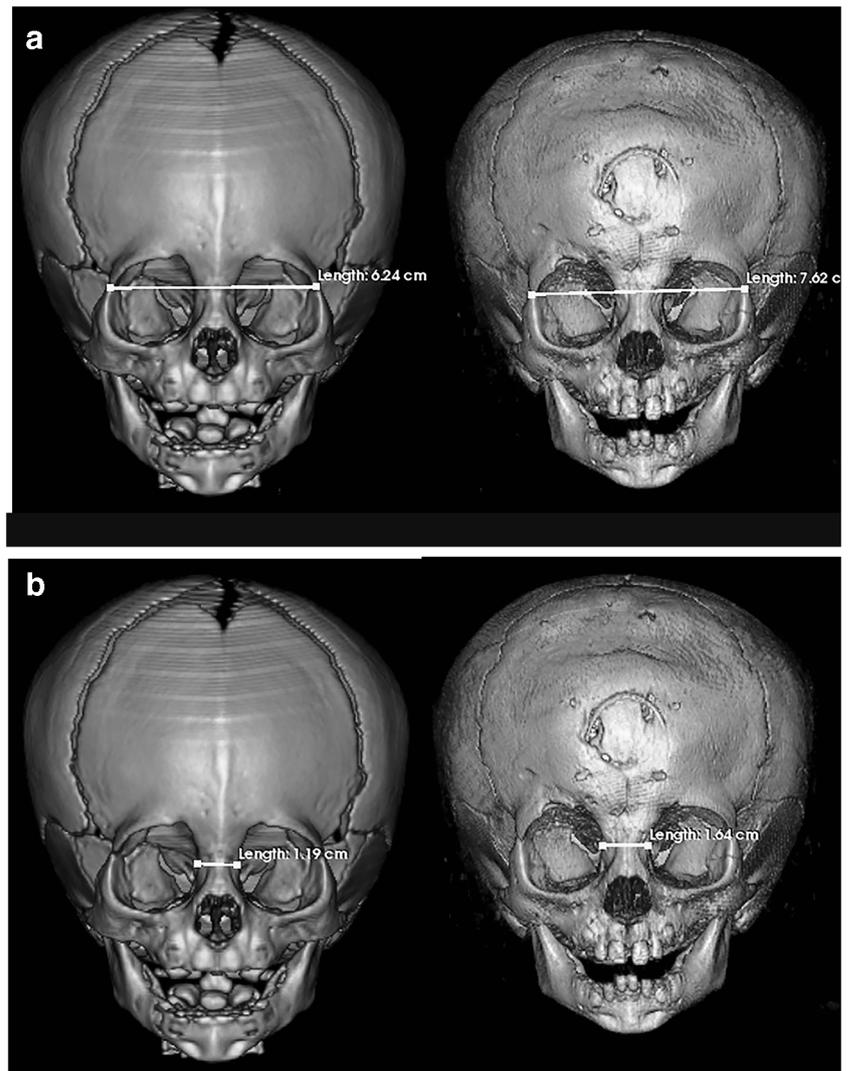
The anthropometric parameters that were followed and compared [2] were as follows (Fig. 4): average change in the



**Fig. 2** (Upper panel) MDCT axial images showing **a** fused internal suture in an infant with trigonocephaly. **b** Open internal suture in a control infant. (Lower panel) MDCT coronal images. **a** Fused nasofrontal and nasal suture in an infant with trigonocephaly. **b** Open

nasal and nasofrontal suture seen in nonmetopic controls. **c** Showing an intraoperative image with Freer dissector showing the open nasofrontal suture in a case of brachycephaly

**Fig. 3 a** MDCT of the infant 3 showing the change in the bony lateral orbital distance, (6.24 to 7.62 cm) at three months follow up. **b** MDCT of the of infant 3, showing the change in the interorbital distance, (from 1.19 cm to 1.64 cm) at three months follow up. (Measurements summarized in Table 1 cf. Normal anthropometry). A strip craniectomy was performed through the fused metopic suture and continued with an osteotome through the nasofrontal suture, confirming that the internasal suture was split. A steel wire spring (length, 12 cm; 17/25 NiTi orthodontic rectangular archwire for the mandibular arch, G&H orthodontics, Langenhagener, Germany) cut to the desired length and shaped as an omega with bayonet-shaped endings at the operating table and was inserted into grooves placed at the edge of craniectomy. One of the springs was placed at the anterior end of the metopic suture close to the nasion. Additional spring is placed higher up frontally to achieve frontal widening and angulation correction



bony lateral orbital distance (LO) achieved at follow-up was 0.88 cm (range 0.45–1.40); average change in interdacyron distance (ID) was 0.24 cm (range 0.07–0.45); average change in intercanthal distance (IC) was 0.30 cm (range 0.10–0.45).

The LO and ID were defined on the 3D reconstruction of the CT and IC on the soft tissue reconstruction (Table 1).

## Discussion

Metopic synostoses present as trigonocephaly or a triangular shaped head. The angulation varies from simple ridging to acute angulation associated with compensatory changes like temporal hollowing and hypotelorism [3, 4].

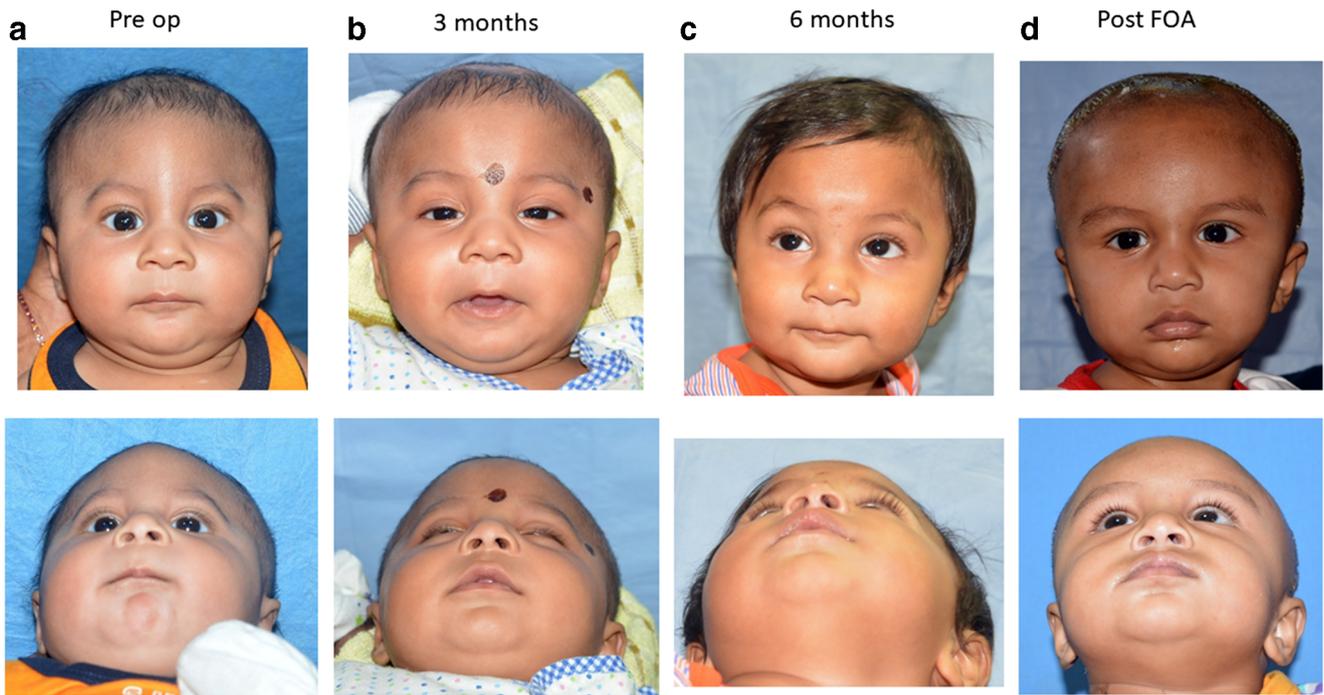
Normal metopic suture usually closes during the first 4–9 months of age, with the earliest evidence of closure, reported at 3 months, initiated at the nasion and completed at the anterior fontanelle [5–8]. Amongst the simple synostoses, metopic synostoses are the only form of synostoses which has a

characteristic angulation at the abnormal suture. The explanation for the angulation in metopic synostoses is unclear from literature.

Our clinicroadiological study demonstrates that the “nasion sutural complex,” i.e., the sutural complex between the paired nasal bone, frontonasal suture line with the possible involvement of frontoethmoid may be responsible for the angulation. Involvement of medial and superior suture lines of the nasal bone, as we have demonstrated radiologically, contributes to the angulation. We did not find any mention in the literature on the nasal bone and its sutural relations and its contribution to trigonocephaly.

**Points corroborating the involvement of nasion sutural complex suture are:**

1. The angulation in trigonocephaly corresponds to the angulation between the nasal bone. The sutural involvement does not allow the “release” of the forehead from the nasal suture and its angulation. It has been well observed



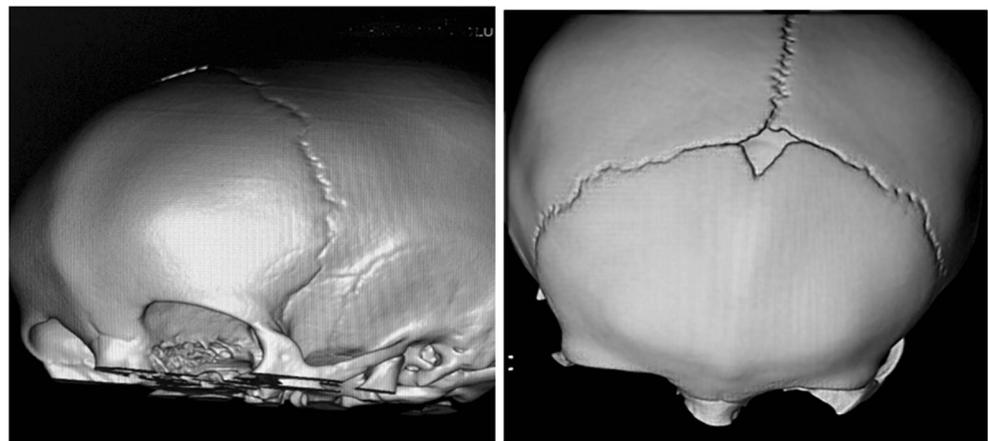
**Fig. 4** The panel is sequentially showing the changes with treatment in an infant (infant 3) with trigonocephaly. The vertical columns in order from left to right **a** The trigonocephalic infant **b** 3 months after spring

cranioplasty **c** 6 months after spring cranioplasty **d** after fronto- orbital correction at nine months along with spring removal

- that the angulation is maximum at the level of the nasal bone and is a marker of the severity of metopic synostoses.
2. The compensatory features in the remaining calvaria in trigonocephaly may be a result of “pinch” at the axial level of the nasal bones [9, 10].
  3. Why do we not see older children and adults with metopic synostoses? [11] We propose that the growth, remodelling and the associated “growth forces” does not allow the angulation to persist, reversing the trigonocephaly.

4. Children with a metopic ridge only have no characteristic features of trigonocephaly. It is kind of metopic involvement belonging to the same spectrum but with no nasofrontal and nasal bone involvement and hence with no consequential trigonocephaly (Fig. 5).
5. The correction of hypotelorism has met with little success with most remodelling techniques. We propose that the hypotelorism is a result of synostosis of the nasion sutural complex with the possible contribution of the frontoethmoid suture accompanying the early fusion of

**Fig. 5** An MDCT of an 8-month-old child with the metopic ridge. Notice the open nasal and nasofrontal sutures contrary to classical trigonocephaly. (Courtesy: Prof Tufan Hicdonmez)



the metopic suture [12] or a consequence of the direct effect of the metopic synostosis over the ethmoid growth. Consequentially, reduction of the medial interorbital wall distance is particularly pronounced compared with that of the lateral orbital walls [13].

In the first 6 to 8 months of life the nasal frontal process, nasal bones, and ethmoid bones are unossified. Considering this fact, this might be an optimal timing for distraction using a spring to reduce the hypotelorism [14, 15] and hence improve correction of trigonocephaly. We believe a satisfactory hypotelorism correction is possible only if the nasion sutural complex is released [16–20]. The release of the sutural complex and applying spring over the glabellar region promotes the sutural distraction and osseous deposition between the orbits correcting the hypotelorism. The compensatory changes may not improve entirely with this technique but can be reduced and may be considered later for revision.

The most important limitation of the study is that it is a small series of patients, and greater numbers will be a key to understand the applicability of the concept of nasion sutural complex to correction of hypotelorism and its long-term effects on trigonocephaly.

## Conclusions

Involvement of nasion sutural complex suture along with the involvement of metopic sutures may explain the angulation in trigonocephaly. We propose that techniques to manage hypotelorism associated with trigonocephaly should release nasion sutural complex for improved results.

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## Compliance with ethical standards

**Conflict of interest** None.

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