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Original article

Sinonasal anatomical analysis of nonsyndromic bilateral coronal suture craniosynostosis using computed tomography

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Abstract

Background: Craniosynostosis is a skull deformity associated with the premature fusion of one or more cranial sutures. In children with syndromic craniosynostosis, anomalies related to the cranial base and midface contribute to midface hypoplasia, which affects the diameter of the axial facial concavity. Nonsyndromic craniosynostosis, particularly bilateral coronal suture craniosynostosis, may result in abnormal facial proportions and affect sinonasal anatomy.

Objective: This study aimed to determine and analyze morphometric measurement values in the sinonasal cavity of patients with nonsyndromic bilateral coronal suture craniosynostosis.

Methods: This retrospective study analyzed 91 children, aged 0 - 10 years, who underwent computed tomography (CT) at King Chulalongkorn Memorial Hospital between January 2010 and November 2022, including 21 children with nonsyndromic bilateral coronal suture craniosynostosis and 70 controls. The following diameters were measured: anterior bony width (ABW), bony choanal aperture width (BCAW), right posterior bony width (RPBW), and left posterior bony width (LPBW). The study group was further divided into four age groups.

Results: The BCAW, RPBW, and LPBW of the nasal cavity in patients with bilateral coronal suture craniosynostosis, aged 0–12 months, were significantly lower than those of the control group. The RPBW and LPBW in the study group, aged > 72 months, were also significantly lower. All measurements were not dependent on children's sex.

Conclusion: This study demonstrated that children with nonsyndromic bilateral coronal craniosynostosis exhibited lower diameters of the bony choanal aperture than healthy children.

Keywords: Children nasal cavity, computed tomography, craniosynostosis.

Craniosynostosis is a condition characterized by the premature closure of cranial sutures, leading to abnormal calvarial growth and resulting in an altered head shape. It may be associated with other malformations, such as the face, body, and extremity, corresponding with syndromes such as Crouzon, Apert, and Pfeiffer (syndromic type), or not associated with syndromes (nonsyndromic type). The nonsyndromic type is more common than the syndromic type. The overall prevalence of craniosynostosis is approximately 1 in 2,500 births, with the nonsyndromic subtype accounting for 0.4 - 1.0 in 1,000 births. [1, 2]

Craniosynostosis disturbs not only calvarial growth but also facial development. In syndromic craniosynostosis, multiple sutures are typically affected, leading to midface hypoplasia characterized by underdeveloped midfacial bones. This feature is commonly observed in various syndromes, including Apert, Crouzon, and Pfeiffer. [1, 2] Midface hypoplasia can result in the retrusion or underdevelopment of the maxilla, contributing to a flat or concave facial profile.[3, 4] Consequently, syndromic craniosynostosis often exhibits more complex and extensive sinonasal abnormalities. For instance, patients with Apert and Crouzon syndromes demonstrate global narrowing of the nasal cavity, whereas those with bilateral choanal atresia exhibit narrowing in the mid- to posterior nasal cavity. [5]

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Nonsyndromic craniosynostosis is typically less complex and usually involves a single suture. However, multiple sutures can be also involved and is characterized as complex nonsyndromic craniosynostosis. Facial development can also be affected by nonsyndromic craniosynostosis, particularly in cases of bilateral coronal suture craniosynostosis, leading to calvarial deformity, specifically brachycephaly, along with midface hypoplasia. [2] The measurement of sinonasal anatomical parameters among these patients is not widely reported, and one study revealed lower diameters of the pyriform aperture and bony choanal aperture in this group. [6]

Details of anatomical changes in the sinonasal region are essential for treatment planning, including surgical treatment and nasal endoscopy. Thus, a comprehensive understanding of paranasal anatomy contributes to achieving optimal outcomes with minimal complications.[4, 5, 7 - 9]

Computed tomography (CT) is the optimal modality for providing detailed and accurate visualization of the craniofacial region, particularly bony structures. This enables precise measurement and assessment of sinonasal structures, offering valuable insights into the details of anatomical changes. Such information is important for effective treatment planning, encompassing both surgical procedures and nasal endoscopy.

This study primarily aimed to determine morphometric measurement values in the sinonasal cavity of patients with bilateral coronal suture craniosynostosis by comparing their CT measurements of the nasal cavity with those of normal children of the same age. The secondary aim was to establish normal ranges of sinonasal measurements in the Thai pediatric population. These CT measurements, together with clinical information, contribute to a better understanding of sinonasal anatomical changes in patients with bilateral coronal sutures craniosynostosis and can be used in treatment planning for patients with nonsyndromic craniosynostosis.

Materials and methods

Study subjects

This retrospective study included children with nonsyndromic bilateral coronal suture craniosynostosis treated at King Chulalongkorn Memorial Hospital (KCMH), Thai Red Cross Society, between January 2010 and November 2022.

The imaging studies of all the patients who underwent standard 3D-CT facial bone imaging with clinical suspicion of craniosynostosis were evaluated. Patients with nonsyndromic bilateral coronal suture craniosynostosis, aged < 10 years, were included. Conversely, those who had syndromic craniosynostosis, history of skull, midface, and sinonasal trauma, unilateral coronal craniosynostosis, and brain and skull base tumors, had undergone post-frontoorbital advancement surgery, post-sinus and midface surgery, and had other congenital anomalies such as facial cleft, midline defect, or frontoethmoidal meningoencephalocele (FEEM) were excluded.

The control group consisted of children aged < 10 years who underwent CT of the facial bone or CT screening of paranasal sinuses for indications unrelated to craniosynostosis without any craniofacial abnormalities on CT. Patients with craniofacial abnormalities, skull fractures, or evidence of sinonasal surgery on CT were excluded. Owing to the small number of patients, all the consecutive cases were included without randomization.

This study was approved by the Research Ethics Review Committee for research involving human subjects.

Data collection

Data were collected from Envision and the Radiologic information system used in KCMH. Demographic information, including sex, age, underlying diseases, and presenting symptoms, was obtained from the hospital information system. Imaging data were independently reviewed on the picture archiving and communication system using Synapse (version 5, Fujifilm Global Japan) by two readers including an experienced, board-certified neuroradiologist and a radiology resident. The average of the two separate measurements was analyzed.

Computed tomography

CT examinations were conducted using the Aquilion S CT scanner, and diagnostic protocols for head CT were adjusted based on age. The parameters were as follows: spiral mode, 80–100 kV tube voltage, and 250 mAs tube current. The reconstructed slice thickness was 1 mm.

3D-CT of the facial bone and screening paranasal sinus were reconstructed in axial view, bone window, parallel to the hard palate, covering the area from the floor of the maxillary sinus to the roof of the frontal sinus.

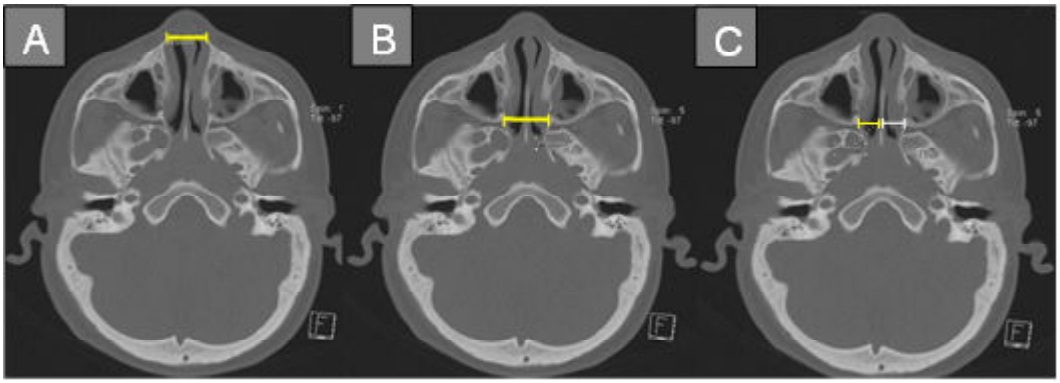

Figure 1. Examples of measurements. **(A)** anterior bony width (ABW); **(B)** bony choanal aperture width (BCAW); **(C)** right and left posterior bony width (RPBW and LPBW).

Nasal cavity measurements

The measurements were conducted twice by a radiology resident and a neuroradiologist directly on CT images, bone window, axial view, and 2D multiplanar reconstruction parallel to the hard palate at the level of the floor of the nasal cavity.

The dimensions of the nasal cavity were measured as follows (**Figure 1**):

- Anterior bony width (ABW) between the two ridges extruding from the maxilla-pyriform aperture
- Bony choanal aperture width (BCAW) between both pterygoid processes and the choanal aperture
- Right posterior bony width (RPBW) between the bone sidewall and septal mucosa
- Left posterior bony width (LPBW) between the bone sidewall and septal mucosa

Statistical analysis

Continuous variables were presented as mean and standard deviation, whereas categorical variables were presented as numbers. The difference between the disease and control groups was analyzed using unpaired Student's *t* - test for continuous variables, with Cohen's d calculated to assess the effect size. Cohen's d values > 0.8 were interpreted as large effect sizes, indicating practical significance. The correlation of nasal cavity diameter in the disease and control groups compared with age was analyzed using linear regression analysis. $P < 0.05$ was considered significant. Statistical analysis was performed using IBM SPSS Statistics for Windows version 28.0 (IBM Corp., Armonk, NY, USA).

Results

Patient characteristics

CT images of 138 patients, who underwent head CT due to clinical suspicion of craniosynostosis, were retrospectively examined. In total, 117 patients were excluded from the study groups, including 23 children without abnormal suture fusion, 15 with genetic syndromes (Apert, Crouzon, and Pfeiffer syndromes), 10 with unilateral coronal suture craniosynostosis, 21 with other craniosynostosis (sagittal, metopic, and lambdoid suture craniosynostosis), 23 with congenital anomalies (FEEM, cleft lip, cleft palate, and facial cleft), and 25 with prior surgery. The remaining 21 patients were included in the study group.

The control group consisted of 70 children who underwent CT of the facial bone or CT screening of the paranasal sinuses without any craniofacial abnormalities or skull fractures.

Both the craniosynostosis and control groups were divided into four age groups: 0 - 12 months (0 - 1 year), 13 - 36 months (>1 - 3 years), 37–72 months (> 3 - 6 years), and > 72 months (> 6 - 10 years) **(Table 1).**

Table 1. Children with bilateral coronal suture craniosynostosis (disease) and those from the control group, divided according to age.

Age	Age	Male		Female		Total	
group	(months)	Disease	Control	Disease	Control	Disease	Control
A	0 - 12	9	6	4	5	13	11
B	13 - 36	3	9	1	3	4	12
C	37 - 72	2	9	-	6	2	15
D	> 72	2	24	-	8	2	32
	Total	**16**	**48**	**5**	**22**	**21**	**70**

Table 2. The anterior bony width (ABW), bony choanal aperture width (BCAW), right posterior bony width (RPBW), and left posterior bony width (LPBW) of the nasal cavity in children with craniosynostosis compared with control group, categorized by sex and age.

Patient's group		ABW (mm) Mean (SD)		BCAW (mm) Mean (SD)		RPBW (mm) Mean (SD)		LPBW (mm) Mean (SD)	
		Disease	Control	Disease	Control	Disease	Control	Disease	Control
	All	13.9 (2.0)	13.5 (0.5)	13.2 (2.7)*	18.2 (0.7)*	6.1 (1.5)*	8.4 (0.5)*	5.8 (1.7)*	8.3 (0.7)*
A	M	13.4 (2.0)	13.4 (0.7)	12.3 (2.3)	18.5 (0.7)	5.6 (1.4)	8.5 (0.2)	5.6 (1.6)	8.5 (0.8)
	F	15.1 (1.5)	13.6 (0.4)	15.4 (2.9)	17.9 (0.6)	7.3 (1.1)	8.3 (0.7)	6.4 (1.8)	8.1 (0.6)
	All	18.9 (2.1)	14.9 (2.0)	18.8 (2.6)	19.9 (1.3)	9.4 (1.8)	9.1 (1.1)	8.7 (1.9)	9.1 (0.9)
B	M	18.1 (1.8)	14.9 (2.1)	19.1 (1.8)	20.1 (1.2)	9.7 (2.1)	9.1 (1.1)	8.7 (1.9)	9.1 (0.8)
	F	21.2 (-)	14.7 (2.6)	18.1 (-)	19.3 (1.6)	8.7 (-)	9.1 (1.2)	7.4 (-)	9.1 (1.4)
	All	17.7 (0.8)	17.2 (1.4)	19.9 (2.2)	23.1 (2.4)	10.3 (1.3)	11.3 (1.3)	10.2 (0.9)	11.2 (1.4)
C	M	17.7 (0.8)	17.2 (1.2)	19.9 (2.2)	22.6 (2.5)	10.3 (1.3)	10.9 (1.1)	10.2 (0.9)	10.8 (1.2)
	F	-	17.2 (1.8)	-	23.8 (2.3)	-	11.9 (1.5)	-	11.9 (1.5)
	All	15.7 (1.4)	17.5 (2.0)	21.4 (1.5)	24.8 (2.3)	10.2 (1.0)*	11.9 (1.4)*	8.6 (0.0)*	12.0 (1.4)*
D	M	15.7 (1.4)	17.0 (2.0)	21.4 (1.5)	24.8 (2.5)	10.2 (1.0)	11.9 (1.4)	8.6 (0.0)	12.1 (1.5)
	F	-	19.0 (1.2)	-	24.5 (2.1)	-	11.9 (1.5)	-	11.8 (1.0)

A (0 - 12 months); **B** (13 - 36 months); **C** (37 - 72 months); and **D** (> 72 months); **F**, females; M, males; and SD, standard deviation. Asterisks (*) indicating significance if the $P < 0.05$.

The detailed results of all measurements are listed in **Table 2**. No significant differences in ABW, BCAW, RPBW, and LPBW were found between boys and girls.

CT parameters of the nasal cavity

The results of the comparative analysis of the nasal cavity dimensions for ABW, BCAW, RPBW, and LPBW in each age group between the patient group and the control group are demonstrated in **Figure 2.**

The BCAW, RPBW, and LPBW of the nasal cavity in the age group 0–12 months were significantly lower than those in the control group ($P < 0.05$). The mean differences (95% confidence interval) in BCAW, RPBW, and LPBW were 5.0 (3.3 - 6.7), 2.3 (1.4 - 3.2), and 2.5 (1.5 - 3.6) mm, respectively. The effect sizes, as measured by Cohen's d, were 2.41, 1.99, and 1.93, respectively, indicating large practical significance.

Significantly lower RPBW and LPBW were measured in the age group > 72 months. The mean differences (95% confidence interval) in RPBW and LPBW were 1.7 (4.1 - 7.6) and 3.4 (2.9 - 3.9) mm, respectively. The effect sizes as measured by Cohen's d were 1.3 and 2.5, respectively, indicating large effect sizes.

The regression model that compared the dimensions of the nasal cavity between the two groups based on age **(Figure 3)** showed a distinct trend, revealing a higher rate of increment in the diameters of the BCAW and RPBW within the older age group with bilateral coronal suture craniosynostosis than in the healthy group. However, the difference in slopes of the graphs between the two groups was not significant (*P*-values for ABW = 0.717, BCAW = 0.102, RPBW = 0.176, LPBW = 0.834).

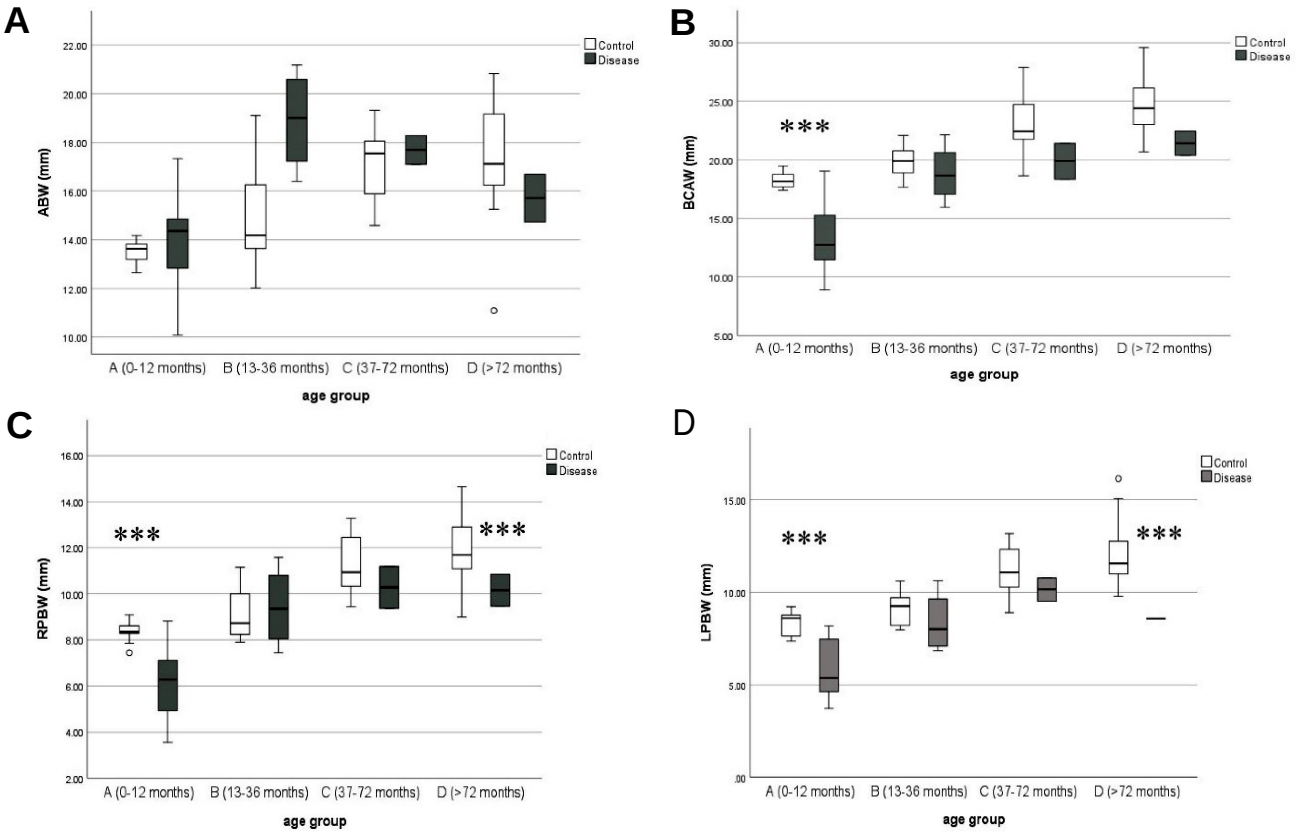

Figure 2. Comparison of healthy controls and bicoronal craniosynostosis patients according to age group in **(A)** anterior bony width, **(B)** bony choanal aperture width, **(C)** right posterior bony width, and **(D)** left posterior bony width. *** indicates $P < 0.05$

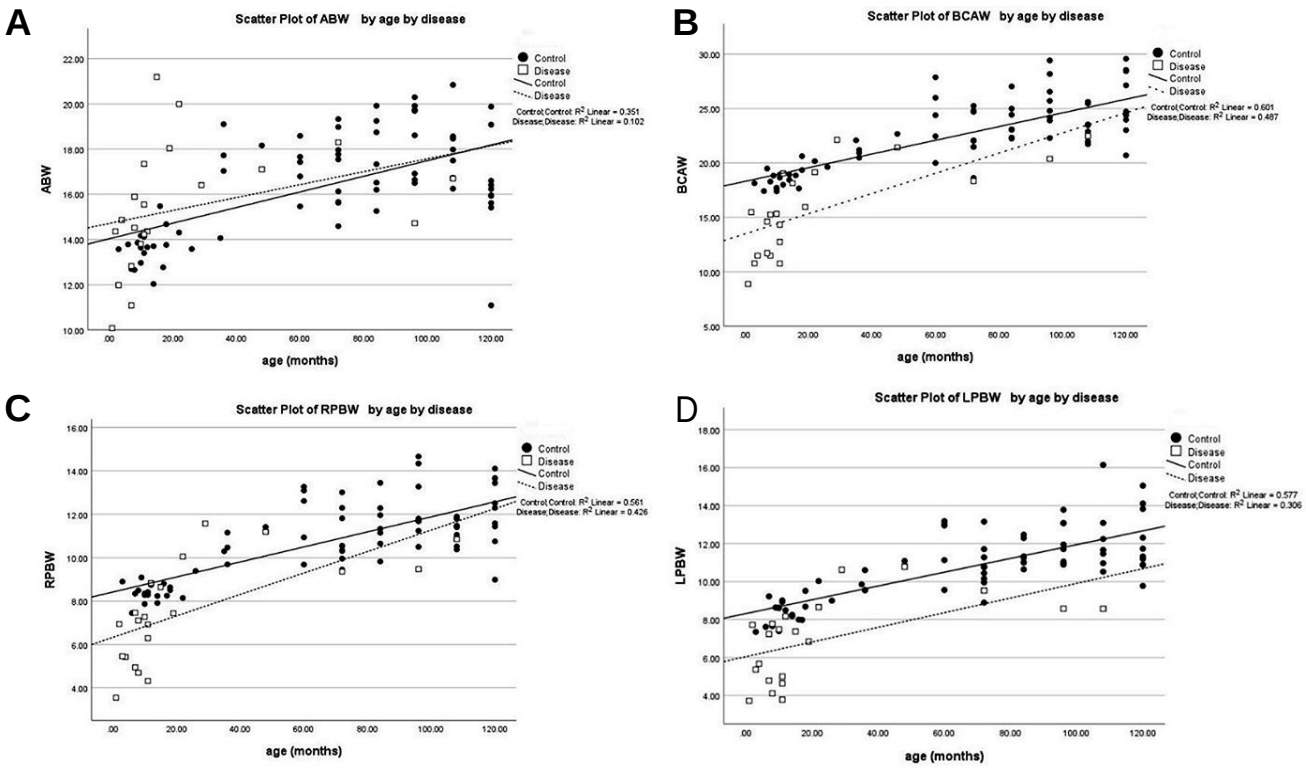

Figure 3. Scattered plots with trend lines show correlation of ABW **(A);** BCAW **(B);** RPBW **(C);** and LPBW **(D)** (mm); and age (month) in disease group (dot line) and control group (line) Anterior bony width (ABW); bony choanal aperture width (BCAW); right posterior bony width (RPBW); and left posterior bony width (LPBW).

Discussion

Craniosynostosis is a cranial deformity characterized by the premature fusion of one or more cranial sutures, often leading to associated facial anomalies, particularly midface hypoplasia, which can result in respiratory difficulties arising from altered nasal anatomy.[2] This condition can be classified into syndromic, which is accompanied by other systemic abnormalities, and nonsyndromic, where craniosynostosis occurs in isolation. The syndromic subtype is more complex and usually involves midface hypoplasia and sinonasal anatomical alterations. Conversely, the nonsyndromic subtype is less complex, and the extent of midface hypoplasia and sinonasal anatomical changes remains not clearly understood. In this study, we mainly investigated and analyzed morphometric measurement values within the sinonasal cavity among patients diagnosed with nonsyndromic bilateral coronal suture craniosynostosis using CT parameters.

CT is widely regarded as the optimal modality for obtaining precise and accurate information about bony structures, including the sinonasal anatomy. It allows detailed measurement and assessment of sinonasal structures, enabling both comparisons with normal individuals and longitudinal follow-up studies. [10] A significant drawback of CT, particularly in children, is the risk of radiation exposure. However, the benefits outweigh this risk, as no other imaging modality can provide comparable details. Currently, CT is the primary modality for preoperative planning in patients with craniosynostosis. Thus, this study sought parameters from CT scans to elucidate anatomical alterations to enhance treatment planning.

In infants who primarily rely on nasal respiration, nasal stenosis can cause respiratory distress. Various conditions, including bilateral choanal atresia, pyriform aperture stenosis, and craniosynostosis, may contribute to nasal obstruction. Graham M, *et al.* (2019) demonstrated that CT measurements revealed narrower dimensions of the entire nasal cavity in patients with pyriform aperture stenosis, Apert syndrome, and Crouzon syndrome than in healthy controls. Moreover, measurements of the posterior half of the nose were also narrower in patients with bilateral choanal atresia. These findings emphasize the significance of considering sinonasal anatomy in cases of craniosynostosis and associated respiratory complications.[5]

Children with syndromic craniosynostosis suffer from anomalies resulting from complex, multiple premature suture fusions, which affect the cranial base and midface, leading to midface hypoplasia and retrusion.[3] Thus, a comprehensive assessment of the midface and nasal cavity before undertaking any surgical intervention or nasal endoscopic procedure is necessary. CT, correlated with clinical findings, has proven beneficial for the diagnosis, treatment planning, and follow-up of Apert and Crouzon syndromes in previous studies. [11-13]

Nonsyndromic craniosynostosis does not correlate with abnormalities in other body parts, such as the face, trunk, and extremities, as observed in syndromic craniosynostosis. The involvement of a single suture is more common than multiple sutures (complex type).[14] Sagittal synostosis is the most common form of isolated craniosynostosis, followed by coronal synostosis and metopic synostosis with the lambdoid synostosis being the least common. [2, 15] Various types of craniosynostosis can have distinct effects on cranial development, necessitating different measurements to comprehensively assess their extent. Bilateral coronal suture craniosynostosis results in skull growth restriction in the anterior–posterior (AP) dimension and widened skull transverse diameter, affecting facial development. This manifests as midface underdevelopment, harlequin deformity, and hypertelorism. Measuring the AP skull length in children could explain the degree of brachycephaly caused by bicoronal craniosynostosis. This study focused on sinonasal structures, which are typically affected in bilateral coronal suture craniosynostosis. Determining the degree and extent of these abnormalities provides valuable information for both clinical aspects and therapeutic planning.

Based on our findings, the nasal apertures were statistically smaller in the group aged 0–12 months with bilateral coronal suture craniosynostosis, with mean differences 95% confidence interval (CI) in BCAW, RPBW, and LPBW of 5.0 (3.3 - 6.7), 2.3 (1.4 - 3.2), and 2.5 (1.5 - 3.6) mm, respectively, all showing large effect sizes. This result confirms the effect and provides insight into how bilateral coronal suture craniosynostosis affects the sinonasal profile. Our results align with those of Gruszczynska K, *et al.* [6], who measured BCAW and found significantly lower values in children with isolated craniosynostosis. In the present study, the craniosynostosis group exhibited smaller posterior

measurement diameters, specifically BCAW, RPBW, and LPBW. This observation may be explained by the likelihood that the pterygoid bones, attached to the skull base, are more susceptible to the disease during the initial year of life. In addition, we found a significantly lower dimensions of RPBW and LPBW in the craniosynostosis group aged > 72 months (6 - 10 years) but not that of BCAW, which could be attributed to differing degrees of nasal septum ossification.

The reduced diameters of the bony choanal aperture in children aged 0 - 12 months with bilateral coronal suture craniosynostosis suggested significant functional and anatomical challenges that may affect nasal airflow, respiratory function, and overall craniofacial development. Midface hypoplasia is a known cause of airway problems in patients with craniosynostosis, leading to breathing difficulty and obstructive sleep apnea, which require tailored treatments specific to individual pathology.[16, 17] This study provides solid evidence of nasal airway narrowing and indicates that CT measurements of the nasal cavity could be valuable in the respiratory care for patients with craniosynostosis, particularly in defining the site and extent of upper airway stenosis caused by midface hypoplasia.

When comparing parameters of children across different age groups, older children with craniosynostosis exhibited a trend of a higher rate of increment than younger ones in the BCAW and RPBW measurements compared with healthy children. This observation supports our hypothesis that the frontoorbital anomaly observed in younger children with bicoronal craniosynostosis may improve over time due to a compensatory mechanism involving the increased widening of the skull base and temporal regions as the children grow. Mercan E, *et al*. [18] measured the growth pattern at the suture sites using preoperative CT images and annotated anatomical landmarks and found that metopic and coronal sutures grew more rapidly in the sagittal craniosynostosis cohort than in the normal cohort. The AP displacement of the semi-landmarks also indicated a more rapid growth in the sagittal plane in the sagittal craniosynostosis model than in the normal model. These findings provide evidence that children with craniosynostosis have different growth rates of the unaffected sutures, which may suggest the compensatory mechanism. However, the slopes of the regression curves in the present study were not significantly different between these two groups, and the LCAW did not display such a trend. This could be attributed to the small sample size or, indeed, anatomical variations in the nasal septum and/or measurement errors. Further studies involving larger populations or longitudinal studies to investigate the progression of sinonasal anatomical changes over time may help validate these results.

Information about cranial growth patterns and timing in patients with craniosynostosis could help in surgical planning when choosing the appropriate age for the operation. The treatment of craniosynostosis focuses on avoiding complications related to premature fusion of the calvarial bones. To plan the most appropriate treatment approach, each case must be carefully analyzed, considering factors such as age, pathology, and clinical manifestation. Simple craniosynostosis can be effectively treated with frontocranial remodeling, whereas syndromic craniosynostosis treatment may involve two stages: addressing cranial vault issues before 6 months and targeting midface retrusion between the ages of 4 and 5. However, new techniques can correct both stenosis and midfacial retrusion in a single surgical procedure such as frontofacial monobloc advancement, yielding good outcomes.[10, 19] We believe that our results could emphasize the effects on the midface and provide valuable information for surgical planning among patients with bilateral coronal sutures craniosynostosis. Moreover, accurate measurements ensure that the surgical correction is precise, potentially reducing the risk of postoperative complications and improving esthetic and functional outcomes.

Sex predilections are observed in craniosynostosis, with certain types showing a sex bias. For instance, in sagittal synostosis, boys are affected more frequently, outnumbering girls in a ratio of 4 : 1. Conversely, unilateral coronal synostosis shows a different trend, with girls outnumbering boys in a ratio of 3 : 2. In metopic, lambdoid, and bilateral coronal synostoses, no clear sex predilection was noted.[2] Among healthy children, different cranial and sinonasal measurements were obtained between sexes. Djupesland P, *et al*. [20], used an optimized acoustic rhinometric probe to measure the nasal airway geometry of patients in their newborn period and at 1 year of age and found that the rhinometric values of male infants were signiûcantly larger than those of female ones. However, these differences were corrected after adjusting for the signiûcantly larger

anthropometric values of male infants, implying that they are related to body size rather than sex. Musa MA, *et al*.[21] examined the head shape of Nigerian children and found relatively higher cranial width in boys than in girls and no significant difference in the cranial length and mean cranial index values between sexes. However, among patients with bilateral coronal suture craniosynostosis included in the present study, no significant differences in the measurement dimensions were found between sexes in both nonsyndromic coronal suture craniosynostosis and control groups.

Apart from the measurement of patients with craniosynostosis, data from healthy controls can be used as a part of normal reference values for the Thai pediatric population, which can serve as a database for future research. These data may be beneficial for understanding other diseases that affect sinonasal anatomy, such as choanal atresia or stenosis, and syndromic craniosynostosis, which can lead to more significant sinonasal anatomical alterations.

This study is limited by the retrospective design, small number of patients, lack of long-term follow-up imaging, and limited clinical information related to the retrospective design. Further research exploring the relationship between morphometric measurements and functional outcomes, such as nasal patency and respiratory efficiency, could enhance the importance of these findings and verify the clinical significance of the abnormal dimensions.

In summary, the reduction of posterior aperture dimensions in children with nonsyndromic bilateral coronal suture craniosynostosis may be associated with the premature fusion of cranial sutures in those aged 0 - 12 months. The results of this study can provide beneficial information for the diagnosis, prognosis, and treatment planning of patients with nonsyndromic craniosynostosis.

Conclusion

This study provides evidence that patients with nonsyndromic bilateral coronal suture craniosynostosis exhibit significant sinonasal anatomical alterations compared with healthy controls. The findings suggest that the pathophysiology of bilateral coronal suture craniosynostosis may involve not only the cranial bones but also the sinonasal structures. These results may help in planning treatment strategies for patients with bilateral coronal suture craniosynostosis. More studies with larger sample sizes and correlation analysis with functional outcomes are needed to validate these findings and fully understand the clinical significance of the sinonasal anatomical changes in bilateral coronal suture craniosynostosis.

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Conflict of interest statement

All authors have completed and submitted the International Committee of Medical Journal Editors Uniform Disclosure Form for Potential Conflicts of Interest. None of the authors disclose any conflict of interest.

Data sharing statement

All data generated or analyzed during the present study are included in this published article. Further details are available for noncommercial purposes from the corresponding author on reasonable request.

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