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Comparison of Black-Bone MRI and 3D-CT in the preoperative evaluation of patients with craniosynostosis

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Abstract

Purpose

Black-Bone (BB) magnetic resonance imaging (MRI) is a non-ionizing imaging method and a recent alternative to computed tomography (CT) in the examination of cranial deformities. The purpose of this study was to compare BB-MRI and routine 3D-CT in the preoperative evaluation of patients with craniosynostosis.

Methods

At our centre we have routinely performed preoperative CT of the skull and brain MRI for patients with clinical suspicion of craniosynostosis. We recently changed our MRI protocol into one that includes sequences for evaluation of both brain anatomy and skull bone and sutures by BB-MRI. A semi-automatic skull segmentation algorithm was developed to facilitate the visualization. Both BB-MRI and 3D-CT were performed on 9 patients with clinical craniosynostosis and the images were evaluated by two craniofacial surgeons, one paediatric neurosurgeon, and two neuroradiologists.

Results

We obtained informative 3D images using BB-MRI. Six (6/9) patients had scaphocephaly, 1 (1/9) patient had unicoronal synostosis, and 2 (2/9) patients had lambdoid synostosis. Affected synostotic sutures could be identified both by BB-MRI and 3D-CT in all patients. Intrarater and interrater reliability for rating the calvarial sutures was high. However, the reliability for rating the intracranial impressions was low by both imaging methods.

Conclusion

BB-MRI is an alternative to 3D-CT in preoperative evaluation of patients with craniosynosytosis.

BB-MRI not only provides information on cranial sutures and intracranial impressions but also on brain structure in one imaging session. This method can replace ionizing radiation-based methods in analysing skull deformities.

Introduction

The coronal, sagittal, lambdoid, and squamosal sutures normally fuse around the fourth decade of life; the metopic suture fuses within the first year of life.[1] Craniosynostosis is defined as the premature fusion of one or more of the sutures between the cranial vault bones.[2,3] These premature fusions can be further classified into categories depending on whether they appear as a part of a syndrome (syndromatic) or as an isolated defect (non-syndromatic).[2,3] The relationship between premature calvarian suture fusion and head shape anomalies in craniosynostosis is established.[3] Craniosynostosis is a relatively rare malformation; the prevalence ranges from 2.6 to 7.2 per 10 000 live births.[4-6] The most common non-syndromatic single suture craniosynostosis is sagittal synostosis, which occurs in 40% to 60% of cases.[4-7]

The diagnosis of craniosynostosis is achieved by physical examination and medical imaging.[2,8] In most cases, the shape of the skull reveals the underlying synostosis. Diagnostic imaging is justified for evaluation of the patency of the remaining sutures and possible intracranial pathology (including Chiari malformation) and to aid in planning the surgical procedure.[2,8-11] Imaging may also reveal signs of diffuse intracranial impressions as a potential indicator of raised intracranial pressure (ICP).[8] In patients with craniosynostosis, imaging has relied on low-dose computed tomography (CT) and on the subsequently constructed 3D images. Despite the unavoidable exposure to ionizing radiation and the well-known risks associated with CT,[12-16] low-dose CT is currently the imaging technique of choice for patients with craniosynostosis.

Children are more sensitive to ionizing radiation than adults.[12,13,17] Thus, whenever available, a non-ionizing imaging modality should be the imaging method of choice. The limitation of MRI has been its poor ability to show bony details. The role of MRI in the diagnostic evaluation of children with skull deformities has thus been minimal. In 2012, Eley et al. presented a novel Black-Bone MRI (BB-MRI) sequence that significantly improved the contrast between soft tissue and bone and allowed visualization of the calvarian bone, sutures, and craniofacial skeleton.[18-21] We introduced the BB-MRI method first for diagnostic imaging of patients with posterior plagiocephaly with neurological symptoms.[22]

The purpose of this study was to compare BB-MRI to CT in the preoperative evaluation of patients with craniosynostosis. 3D-reconstructed images of the cranial vault were created from the data obtained from the two imaging modalities and the appearance of the cranial sutures was assessed. We also evaluated the quality of the 3D reconstructions created with BB-MRI and compared them with 3D reconstructions presented in the literature. Our aim was to provide high-quality 3D-reconstructed images of the cranial vault for routine clinical practice with a non-ionizing imaging method.

Patients and methods

Ethical approval for the use of BB-MRI was granted from the Helsinki University Hospital Research Ethics Committee. It is a routine practice at our craniofacial centre to perform MRI of the brain as a preoperative evaluation of potential intracranial findings (such as Chiari malformation) for all patients with craniosynostosis. From 2016 to 2018 a total of 9 children with clinical suspicion of craniosynostosis underwent 3D-CT and an MRI protocol that included sequences for evaluation of both brain anatomy and skull bone and sutures by BB-MRI. The accuracy and diagnostic value of BB-MRI to confirm the diagnosis of craniosynostosis and to visualize synostotic or patent sutures and intracranial impressions was tested by comparing the imaging data received by

3D-CT and BB-MRI. In this study population, BB-MRI imaging was performed in the same imaging session with routine brain MRI.

MRI was performed with a Siemens Skyra 3T (Erlangen, Germany) with a 32-channel head coil. Two MRI protocols were set up for imaging depending on patient age and need for anaesthesia. The feed-and-sleep method was used for patients under 6 months and general anaesthesia for others. Patients P2, P4, and P5 were imaged using the bottle-fed protocol, which utilizes Siemens quiet suite sequences and consists of an in-phase Blackbone StarVibe sequence and T2-weighted quiet BLADE coronal and axial sequences. The StarVibe sequence employs a stack-of-stars k-space acquisition, which is less motion-sensitive than the VIBE sequence.[23] The remaining patients were imaged under general anaesthesia and the imaging protocol is presented in Kuusela et al.[22] The image segmentation for the Blackbone Starvibe sequence was performed by altering the algorithm presented in Kuusela et al. to use only the inphase image.[22] For the StarVibe sequence, large parts of the skull had to be segmented manually. The development of the sequence and algorithm is ongoing.

Image analyses

Radiological evaluation of the structural and BB-MRI sequences was performed by two neuroradiologists (NB, PV). Information on the cranial sutures and intracranial impressions received from 3D-CT and BB-MRI was evaluated by two craniofacial surgeons (JL, AS), one paediatric neurosurgeon (AK), and two neuroradiologists (NB, PV). Interrater reliability between raters was calculated. The two neuroradiologists performed the evaluation blinded and repeated the evaluation after 2 weeks to evaluate intrarater reliability. Five (sagittal, right coronal, left coronal, right lambdoid, and left lambdoid) sutures of each patient were included in the analyses. Anterior and posterior sagittal sutures were rated separately. Each of the sutures were rated to be open, partially fused, or completely fused. Impressions were evaluated in three different locations of the skull: anterior, parietal, and posterior (see Table 3). In each location, intracranial impressions were graded as severe (2), mild (1), or absent (0).

Statistical analyses

Intrarater reliability was calculated both with percent agreement and Cohen's κ between ratings. Interrater reliability between the five raters was calculated by Kripperndorff's Alpha. Analyses were performed using Microsoft Excel and SPSS version 24.

Results

This study material consisted of 9 patients (5 females and 4 males) with clinical diagnosis of craniosynostosis and clinical indication for preoperative CT and MRI. Six (6/9) patients had scaphocephaly, 1 (1/9) patient had unicoronal synostosis, and 2 (2/9) patients had lambdoid synostosis. Diagnosis of all patients was confirmed by routine 3D-CT. The average age at the time of CT was 27.8 (range 1.6-91.6) months and at the time of BB-MRI 29.5 (range 2.6-94.8) months (see **Table 1**).

We obtained informative 3D images of the skull bone and sutures by using BB-MRI (**Figures 1-4**). The skull segmentation was based on bias field corrected fuzzy c-means clustering,[24] which aims to calculate probabilities of belonging to a cluster. This requires that the surrounding of a voxel consists of similar types of voxels to be classified as belonging to a cluster. The slice thickness of the BB-MRI sequence is 1 mm and the skull of infants is approximately the same thickness. Thus, classifying the clusters correctly in infants with thin skull was more challenging.

The imaging data obtained by routine 3D-CT and BB-MRI were compared by all reviewers (see summary in **Table 2**). The same diagnosis of craniosynostosis was made for 9 patients by all five reviewers. While the calvarian sutures were also visualized from the BB-MRI images in the youngest (2-5 months) patients **(see Figure 4**), the visualization of the bony structures was more accurate in the older patients. In patient number P8 with sagittal synostosis, the right coronal suture was also rated partially synostotic in BB-MRI by the paediatric neurosurgeon. In patient number P4 with sagittal synostosis, the right lambdoid suture was also rated to be partially synostostotic by one of our neuroradiologists. Interrater reliability for evaluating the sutures by routine 3D-CT and BB-MRI were Kalpha 0.953 and Kalpha 0.950, respectively. The intrarater reliability between ratings by routine 3D-CT with percent agreement was 0.963 (neuroradiologist 1) and 1.00 (neuroradiologist 2). The Cohen's κ reliability was found to be κ=0.914 (neuroradiologist 1) and κ=1.00 (neuroradiologist 2). The intrarater reliability between ratings by BB-MRI with percent agreement was 0.907 (neuroradiologist 1) and 0.870 (neuroradiologist 2). The Cohen's κ reliability was κ=0.8 (neuroradiologist 1) and κ=0.731 (neuroradiologist 2).

Intracranial impressions were evaluated by the reviewers in 3D-CT and BB-MRI; the results are summarized in **Table 3**. A variable amount of intracranial impressions was detected in the skull bone in all 9 patients. There was some variation in how impressions were visualized by 3D-CT in comparison to BB-MRI (Table 3). The impressions were typically rated more intense in BB-MRI in comparison to 3D-CT (5/27). In one patient, 1/27 impressions were rated more intense in 3D-CT in comparison to BB-MRI. In most comparisons, the impressions were evaluated similarly in BB-MRI and 3D-CT (21/27). Interrater reliability for assessment of impressions was low by both methods (3D-CT, Kalpha 0.553; BB-MRI, Kalpha 0.458). Intrarater reliability between ratings by routine 3D-CT with percent agreement was 0.667 (neuroradiologist 1) and 0.778 (neuroradiologist 2). The

Cohen's κ reliability was found to be κ=0.461 (neuroradiologist 1) and κ=0.634 (neuroradiologist 2). Intrarater reliability between ratings by BB-MRI with percent agreement was 0.778 (neuroradiologist 1) and 0.667 (neuroradiologist 2). The Cohen's κ reliability was κ=0.62 (neuroradiologist 1) and κ=0.491 (neuroradiologist 2).

In our study, no structural malformations of the brain were observed in any patients. Patient number P8 had two very small left cerebellar T2 dark signal foci in a location typical for perinatal germinal matrix haemorrhage. The visualization of the bony structures with BB-MRI was found to be more accurate in older patients compared to patients under 6 months.

Discussion

The aim of this study was to assess BB-MRI in the preoperative evaluation of patients with craniosynostosis. The BB-MRI method presented by Eley et al.[19] was used and further developed as a suitable MRI protocol for diagnostic imaging of patients with craniosynostosis.

In routine diagnostic evaluation of children with skull deformities, CT allows documentation of the patency or closure of the sutures and aids in surgical planning.[2,8] Most cranial sutures are best assessed using 3D reconstruction of images, as these provide information that is not revealed on axial 2D-CT.[25] However, the potential risks associated with exposure to radiation are well known and thus some authors argue that 3D-CT should not be routinely used in craniosynostosis.[9,12,16] The results of the current study show that the BB-MRI protocol also provides high-quality 3D-reconstructed skull images and visualisation of calvarian sutures and skull bone structure. Preoperative findings and diagnostic assessment on BB-MRI were consistent with findings from 3D-CT in 9 patients with single-suture synostosis. As a limitation of this study, the number of the patients in this series is relatively low. However, all images were analysed by five raters and all affected synostotic sutures could be identified by each rater from both BB-MRI and 3D-CT images, which increases the power of this study.

Raised ICP is one of the most serious functional concerns in craniosynostosis. The presence of diffuse intracranial impressions (or copper beating) in the inner calvarian surface and erosion of the dorsum sella are typical signs associated with increased intracranial pressure.[26] Whether mild or localized intracranial impressions represent raised ICP is, however, controversial.[26] To a certain degree, intracranial impressions are considered as a "normal" finding in a young child with active growth of cranium.[27] In patients with sagittal synostosis, a high incidence of intracranial impressions has been reported[28] and "diffuse" intracranial impressions are considered a specific sign for raised ICP.[26] 3D-CT has been used to identify intracranial impressions in the skull bone. Based on the current findings, both 3D-CT and BB-MRI seem to be subjective methods for evaluating the extent of calvarial impressions. Intrarater reliability for both techniques were also relatively low for this purpose. MRI seems to slightly overestimate the extent and severity of the impressions compared to CT, but it is unclear which of the methods better estimate the true intraoperative situation.

We observed that image segmentation of the skull is more difficult in younger patients. The skull of a 6-month-old or younger infant is quite thin and has a higher water content and thus the bone is not always black. In the current study, the calvarian sutures were also visualized from the BB-MRI images in the youngest patients, but the visualization of the bony structures was more accurate in the older patients compared to the patients under 6 months. While MRI eliminates the risks of ionizing radiation, the increased examination time, need for anaesthesia, lower availability, and cost may be disadvantages of BB-MRI. Of all patients who receive sedation/anaesthesia for diagnostic procedures the pediatric population represents the highest risk and lowest error tolerance subgroup.[29] Especially for infants, the greater anaesthesia or sedation time can be considered a potential risk for the developing brains.[30] The black-bone sequence itself lasts approximately 3 minutes, but if there is need to image brain, the whole protocol will take approximately 15 minutes. In this study, BB-MRI was performed in natural sleep after feeding for patients under 6 months and a less motion-sensitive MRI sequence was used. For patients with scaphocephaly, we aim to perform cranioplasty between the age of 4 to 6 months, thus imaging at infant age is indicated but it is possible without anaesthesia.

Another radiation-free modality for evaluation of cranial sutures that does not require anaesthesia is cranial ultrasound.[31] However, this approach has not yet gained widespread acceptance, perhaps because it is age-dependent and children with a positive finding at sonography are still often recommended to undergo CT for more precise preoperative evaluation and surgical planning.

MRI is superior to CT in most cases when evaluating structural brain alterations in children. MRI is routinely used in patients with craniosynostosis and some neurological abbreviations. A coexistent Chiari malformation is found in some children with single-suture and frequently with syndromic synostosis.[32,33] In such instances, a tailored and more posterior cranial remodelling may be indicated. One major advantage of BB-MRI over CT is avoidance of harmful radiation exposure.[12] Another advantage is that information of the skull bone, sutures, and brain can be obtained in one imaging session. Of our 9 patients with single-suture synostosis, only one had minor cerebellar germinal matrix haemorrhages considered as an incidental finding.

In syndromic craniosynostosis, there is a high risk of concomitant dural venous abnormalities due to abnormal dural sinus maturation, venous hypertension, and veno-occlusive disease.[34] For these patients, venous imaging is needed for preoperative planning. Knowledge of the exact location, shape, and size of venous sinuses is very useful information for the surgeon in all types of cranial remodelling procedures, including both single-suture and syndromic craniosynostosis. The risk of venous sinus tear should be minimized while making the bony cuts over the midline. We believe that MRI may have an important role in preventing this major complication. MR venography provides information on the extent of veno-occlusive disease and on the brain. In the future, combining the BB sequence with MRI venography and routine brain MRI will cover all critical aspects in preoperative planning needed for complex syndromic craniosynostosis cases.

In conclusion, preoperative findings and diagnostic assessment on BB-MRI were consistent with findings on the 3D-CT in 9 patients with single-suture synostosis. Thus, the MRI protocol with the BB-MRI sequence used in this study provides a very promising alternative to CT when imaging patients with craniosynostosis or any calvarial deformity. This protocol provides information on all critical aspects needed for preoperative planning in patients with craniosynostosis.

Author contributions

Saarikko Anne: study design, collecting patient material, analysis of patient material, writing

Mellanen Eero: writing, analysis of patient material

Kuusela Linda: writing, development of imaging protocol, analysis of patient material

Leikola Junnu: collecting patient material, analysis of patient material

Karppinen Atte: analysis of patient material, writing

Autti Taina: study design, writing

Virtanen Pekka: neuroradiologist, analysis of patient material

Brandstack Nina: neuroadiologist, analysis of patient material, writing

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Conflicts of interest: None declared
Ethical approval: Ethical approval for the use of BB-MRI was granted from the Helsinki University Hospital Research Ethics Committee.

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Figure Legends

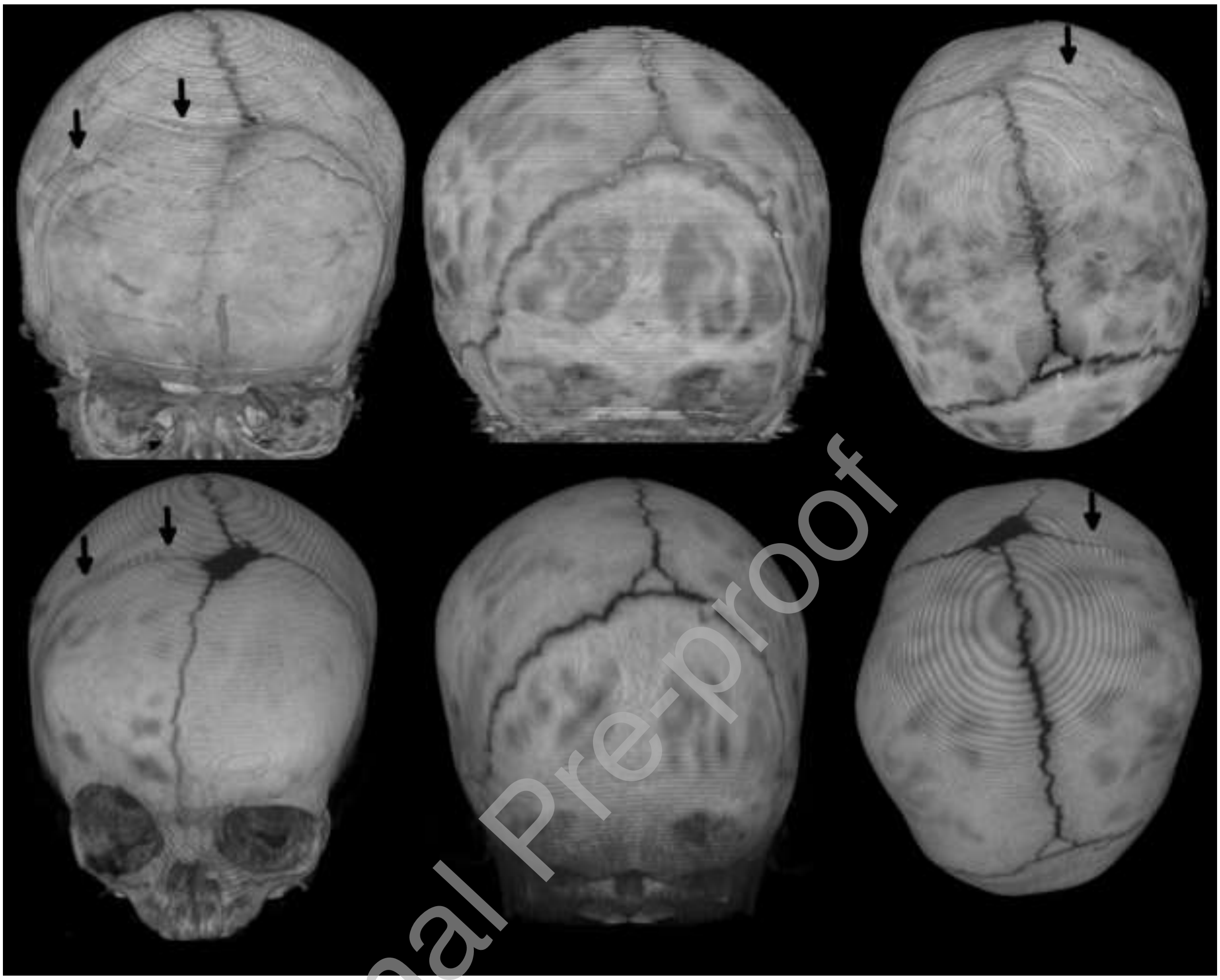

Figure 1. Skull images of patient 1 with right coronal synostosis. Top row, 3D-rendered BB-MRIs at the age of 7 months. Bottom row, 3D-rendered CT images of the same patient at the age of 6 months. **The arrows point to the location of the fused suture.**

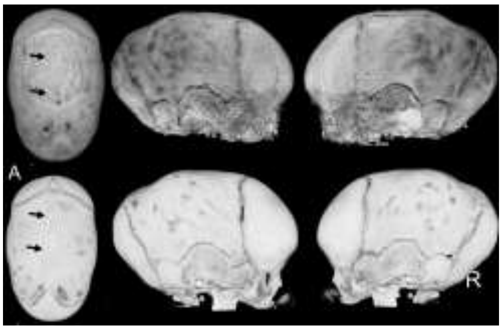

Figure 2 Skull images of patient 5 with sagittal synostosis. Top row, 3D-rendered BB-MRIs at the age of 4 months. Bottom row, 3D-rendered CT images at the age of 5 months. **The arrows point to the location of the fused suture.**

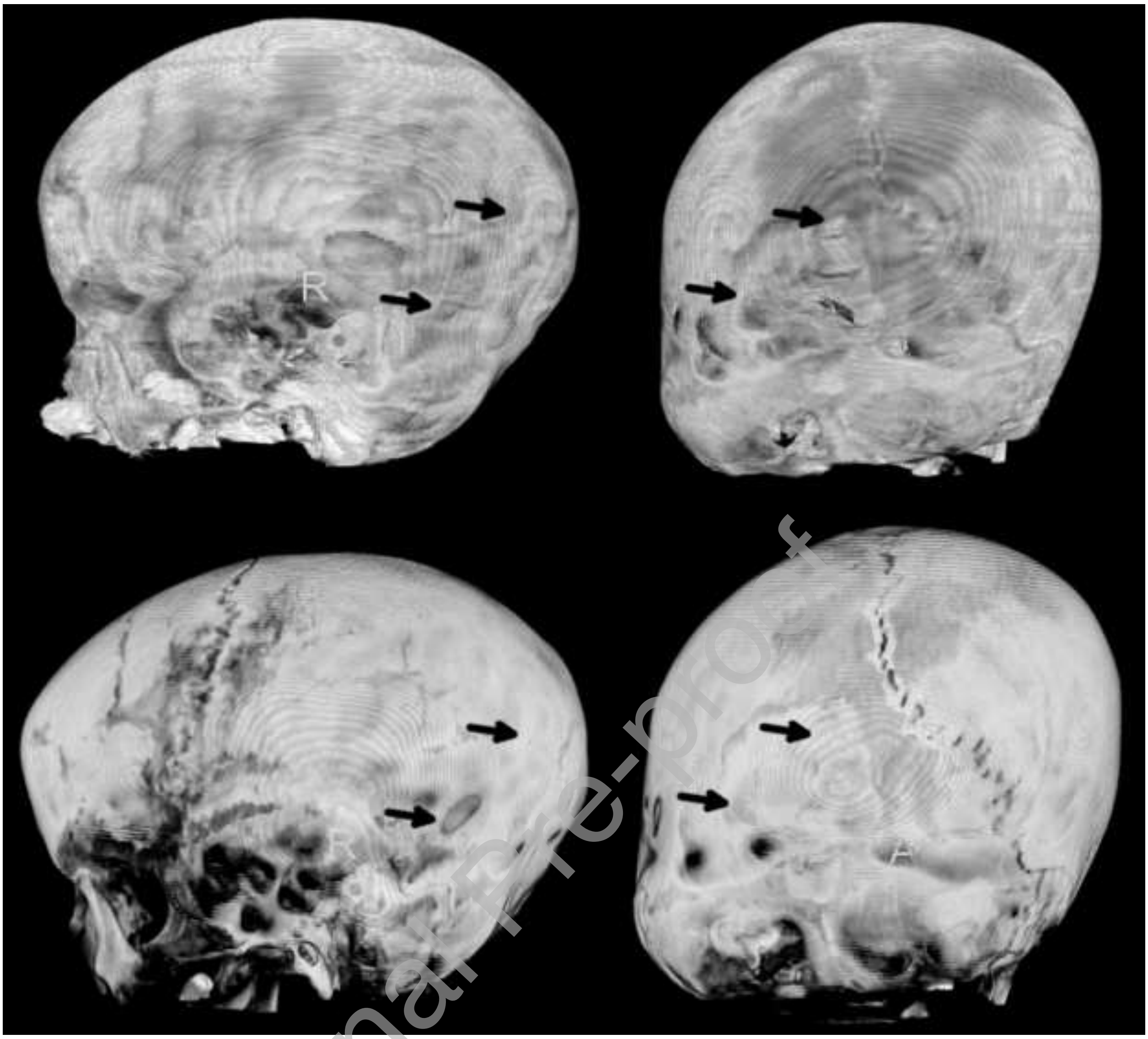

Figure 3. Skull images of patient 7 with left lambdoid synostosis. Top row, 3D-rendered BB-MRIs at the age of 78 months. Bottom row, 3D-rendered CT images at the age of 76 months. **The arrows point to the location of the fused suture.**

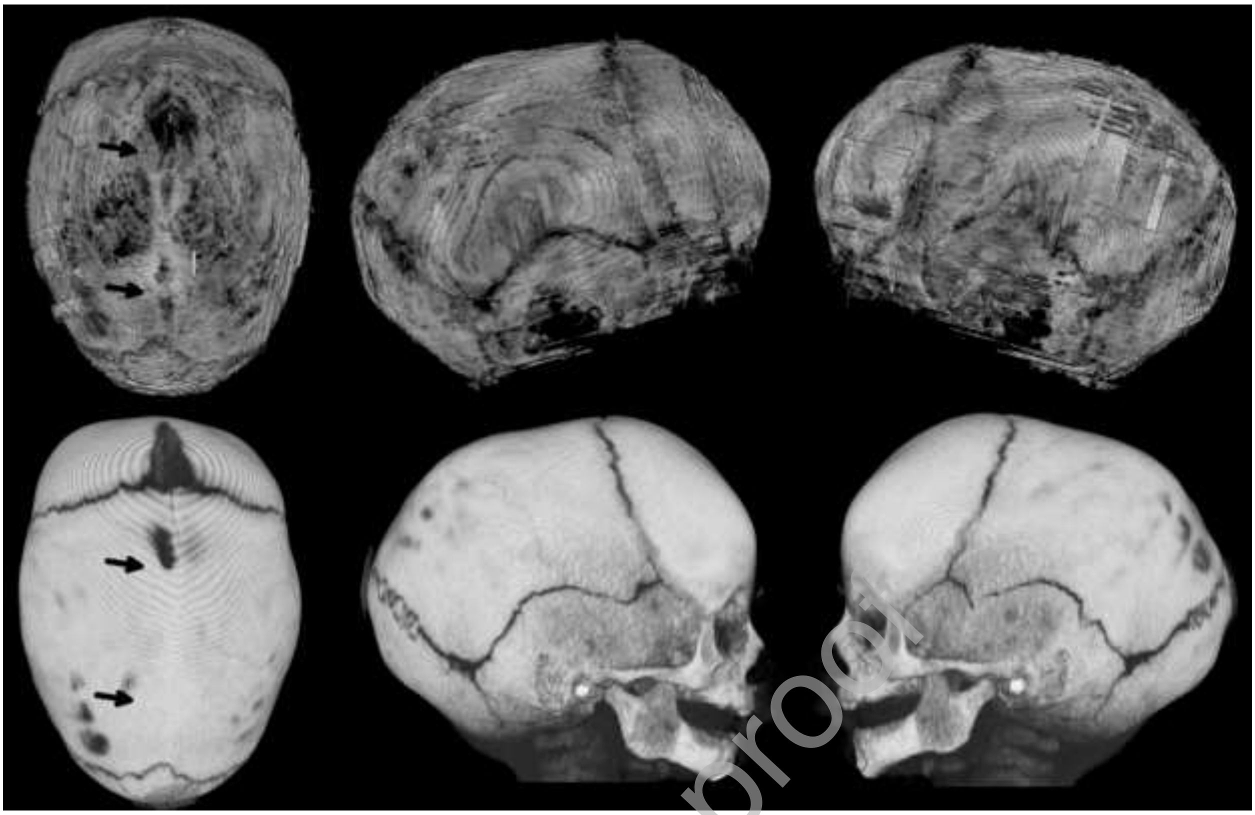

Figure 4. Skull images of patient 3 with sagittal synostosis. Top row, 3D-rendered BB-MRIs at the age of 2.6 months. Bottom row, 3D-rendered CT images at the age of 2.8 months. Imaging was performed in natural sleep after feeding. **The arrows point to the location of the fused suture.**

Table 1.

Patient ID	Diagnosis/synostosis	Sex	Age in CT (M)	Age in BB-MRI (M)
P1	Unicoronal synostosis, right	F	6.2	7.9
P2	Sagittal synostosis	M	5.6	4.9
P3	Sagittal synostosis	M	2.8	2.6
P4	Sagittal synostosis	F	55.9	59.9
P5	Sagittal synostosis	M	5.1	4.1
P6	**Unilamboid** synostosis, left	F	1.6	7.5
P7	**Unilamboid** synostosis, left	F	76.2	78.9
P8	Sagittal synostosis	F	5	5.2
P9	Sagittal synostosis	M	91.6	94.8

TABLE 2.

ID	Diagnosis by CT	Diagnosis by BB-MRI						
		Craniofacial surgeon 1	Craniofacial surgeon 2	Neurosurgeon	Radiologist 1	Radiologist 1, 2nd evaluation	Radiologist 2	Radiologist 2, 2nd evaluation
P1	Right coronal synostosis	Right coronal	Right coronal	Right coronal	Right coronal	Right coronal	Right coronal	Right coronal
P2	Sagittal synostosis	Sagittal	Sagittal posterior*	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal
P3	Sagittal synostosis	Sagittal posterior*	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal
P4	Sagittal synostosis	Sagittal	Sagittal	Sagittal	Sagittal and right lamboid*	Sagittal, right lamboid* and left coronal*	Sagittal	Sagittal and left coronal*
P5	Sagittal synostosis	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal
P6	Left lamboid synostosis	Left lamboid	Left lamboid	Left lamboid	Left lamboid	Left lamboid	Left lamboid	Left lamboid
P7	Left lamboid synostosis	Left lamboid	Left lamboid	Left lamboid	Left lamboid	Left lamboid	Left lamboid	Left lamboid
P8	Sagittal synostosis	Sagittal	Sagittal	Sagittal and right coronal*	Sagittal	Sagittal	Sagittal	Sagittal and right coronal*
P9	Sagittal synostosis	Sagittal	Sagittal	Sagittal	Sagittal	Sagittal and left lamboid*	Sagittal	Sagittal

Table 3. Impressions – CT vs BB-MRI.

	P1		P2		P3		P4		P5		P6		P7		P8		P9	
	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI	CT	BB-MRI
Craniofacial surgeon 1	011	011	011	011	011	011	011	010	011	012	---	012	001	001	012	012	---	001
Craniofacial surgeon 2	021	022	012	012	012	012	112	112	012	012	---	012	001	001	012	012	---	111
Neurosurgeon	111	011	001	011	011	011	001	011	010	011	000	011	011	011	010	000	011	000
Radiologist 1	122	122	011	121	021	121	010	010	011	022	000	021	000	111	021	021	000	000
Radiologist 1*	122	122	021	011	022	011	000	010	021	022	000	022	000	011	021	011	011	011
Radiologist 2	122	122	011	011	021	011	010	011	021	022	000	022	111	121	011	111	010	111
Radiologist 2*	122	121	121	011	122	111	011	121	121	022	---	022	111	122	021	211	011	111

*Second evaluation.