

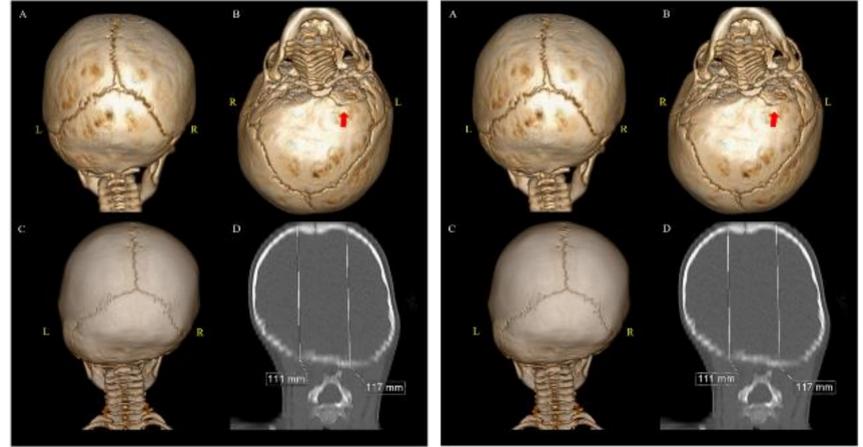
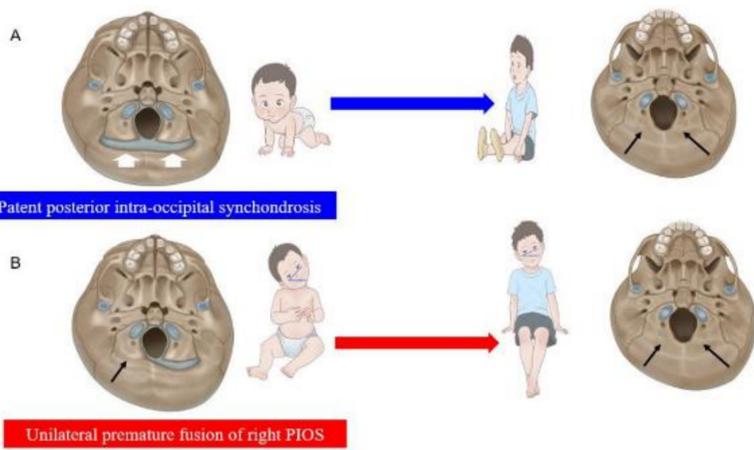
New Insights into the Relationship Between Skull Base Fusion and Torticollis in Children

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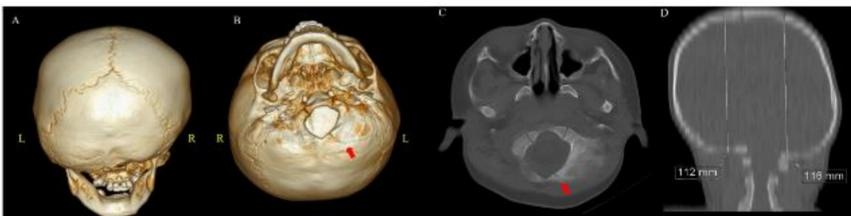
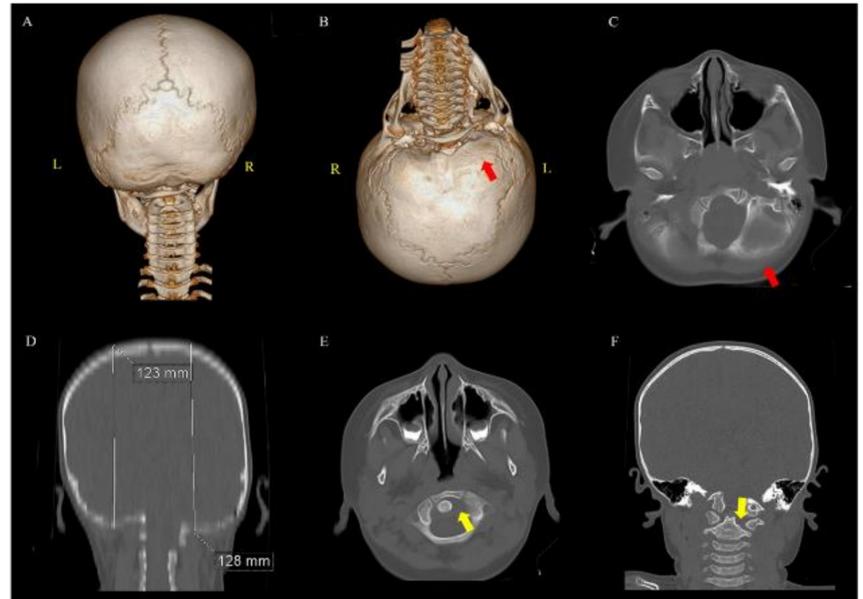
Objective: This study investigates the connection between unilateral premature fusion of the posterior intra-occipital synchondrosis (UPF-PIOS) and head tilt in pediatric patients. We aimed to analyze the clinical features of children diagnosed with UPF-PIOS who also exhibit head tilt.

Table 2. Contingency table showing the relationship between the side of unilateral premature fusion of the posterior intra-occipital synchondrosis (UPF-PIOS) and the side of head tilt.

Side of UPF-PIOS	Side of head tilt		p-value
	Right	Left	
Right	6	0	0.0002
Left	0	9	



Study design: From January 2005 to July 2024, we evaluated patients visiting the Center for Torticollis due to torticollis. UPF-PIOS was diagnosed based on three criteria: (1) asymmetry of the posterior skull base identified through visual assessment of 3D skull images, (2) confirmation of UPF-PIOS on 3D or axial craniofacial CT images, and (3) an absolute difference of 2 mm or more in the vertical length of the skull between the right and left sides on coronal CT images.



Results: Among 1,680 patients, we identified 15 with UPF-PIOS (0.89%). Females predominated (12 girls, 3 boys, $p = 0.035$). The median age at diagnosis was 19 months, with most patients diagnosed before age 3. The median vertical length difference between the right and left sides of the skull was 8.0 mm, with the affected side shorter. All patients exhibited head tilt toward the side of UPF-PIOS, and 53% had craniovertebral junction abnormalities.

Conclusions: We found UPF-PIOS to be a significant cause of torticollis in children. Four key clinical features of UPF-PIOS include (1) presence of UPF-PIOS, (2) asymmetric posterior skull base, (3) shorter skull on the affected side, and (4) head tilt toward the UPF-PIOS side. This consistent observation suggests UPF-PIOS may be an underrecognized cause of torticollis, warranting consideration in differential diagnoses. Further research with larger groups is needed to confirm these findings.

Table 1. The characteristics of the patients with unilateral premature fusion of the posterior intra-occipital synchondrosis (UPF-PIOS)

Characteristics	Values
Number of patients	15
Number of boys to girls (%)	3:12 (20:80)
Age at the time of computed tomography scan (median, interquartile range, months)	19.0 (12.2 -28.5)
Absolute value of maximal vertical length difference between the right and left sides of the skull (median, interquartile range, mm)	8.0 (6.0-9.5)
Side of UPF-PIOS, right to left (%)	6:9 (40:60)
Side of head tilt, right to left (%)	6:9 (40:60)
Number of patients with head tilt toward shorter side of the skull (%)	15 (100)
Presence of craniovertebral junction abnormalities, yes to no (%)	8:7 (53:47)

