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Functional Outcome of Surgically Treated Cases of Primary Craniosynostosis

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ABSTRACT

Background: *Craniosynostosis is a congenital condition characterized by premature fusion of one or more cranial sutures, potentially impairing brain development. Early surgical correction is widely recommended, but the influence of specific variables such as age at surgery, number and type of affected sutures, and surgical technique on long-term functional outcomes remains under investigation.*

Objective: *To evaluate the relationship between functional outcomes and (1) age at the time of surgery, (2) number and type of affected cranial sutures, and (3) type of surgical intervention in patients with primary non-syndromic craniosynostosis.*

Methods: *This retrospective analytical study included 58 children with primary non-syndromic craniosynostosis who underwent surgical treatment at El Sahel Teaching Hospital between July 2009 and July 2020. Patients were assessed five years postoperatively using the Stanford-Binet Intelligence Scales, Fifth Edition (SB5), and the Assessment and Programming of Psycho-Motor Skills (APPS) to evaluate cognitive and psychomotor outcomes, respectively. Statistical analysis was performed using SPSS v25.0, with significance set at $P < 0.05$.*

Results: *The mean age at surgery was 6.79 ± 4.9 months. A strong negative correlation was found between age at surgery and SB5 score ($r = –0.521$, $P < 0.001$), indicating better cognitive outcomes with earlier intervention. Patients operated upon at ≤6 months had significantly higher SB5 scores compared to those treated at older ages ($P < 0.001$). APPS scores were also higher in younger age groups, though the difference was not statistically significant ($P = 0.278$). A greater number of affected sutures was associated with poorer outcomes in both SB5 and APPS scores ($P < 0.001$). Patients with unicoronal or metopic synostosis had better outcomes, while pansynostosis and metopic-bicoronal involvement were linked to lower scores. Suturectomy was associated with significantly better outcomes than reconstructive surgery (SB5: $P < 0.001$; APPS: $P = 0.04$).*

Conclusion: *Early surgical intervention, fewer affected sutures, and suturectomy were all associated with improved cognitive and psychomotor outcomes in patients with non-syndromic craniosynostosis. These findings support the prioritization of early diagnosis and minimally invasive surgical approaches to optimize long-term neurodevelopmental outcomes. Adoption of standardized neurodevelopmental tools such as the Stanford-Binet Intelligence Scales and the APPS test is recommended to evaluate cognitive and psychomotor outcomes of such cases periodically before and after the procedure.*

Keywords
Craniosynostosis, Neurodevelopment, Early surgery, Stanford-Binet, Suturectomy, APPS, Cognitive outcome, Psychomotor function.

Abbreviations
ICP: Increased intracranial pressure, CT: Computed Tomography, 3D: Three-dimensional reconstruction, QOL: Quality of life, SB5: Stanford-Binet Intelligence Scales, Fifth Edition, APPS: Assessment and Programming of Psychomotor Skills.

Introduction
Craniosynostosis is a congenital cranial deformity characterized by the premature fusion or growth arrest of one or more cranial sutures, resulting in abnormal skull morphology. In more severe presentations, infants may develop increased intracranial pressure (ICP), facial dysmorphism, and respiratory or neurocognitive impairments. The etiology of craniosynostosis is multifactorial. While approximately 20% of cases are associated with identifiable genetic mutations, often inherited in an autosomal dominant pattern, the majority are of unknown (sporadic) origin, potentially influenced by maternal and environmental factors. Nonsyndromic craniosynostosis accounts for nearly 75% of cases and typically involves isolated suture fusion without systemic anomalies. The remaining cases are syndromic, frequently associated with well-characterized genetic syndromes such as Crouzon, Apert, and Pfeiffer syndromes, among over 180 recognized syndromes linked to craniosynostosis [1-3].

The estimated prevalence of craniosynostosis ranges from 1 in 2,000 to 2,500 live births. While diagnosis is primarily clinical, often based on abnormal skull shape and growth patterns within the first year of life, imaging studies play a key role in confirmation and surgical planning. Skull radiographs may provide initial clues, but cranial computed tomography (CT), particularly with 3D reconstruction, remains the gold standard for accurately identifying fused sutures and assessing cranial morphology prior to surgical intervention [4,5].

If left untreated, craniosynostosis can significantly impair a child's neurocognitive development, alter craniofacial aesthetics, and reduce overall quality of life (QOL). Delayed intervention may result in persistently elevated intracranial pressure, developmental delays, and psychosocial consequences related to facial asymmetry and stigmatization [6,7].

Surgical intervention remains the standard of care for nonsyndromic craniosynostosis, aiming to prevent the progression of skull deformity and potential neurodevelopmental compromise. However, in select mild cases, some families may choose conservative (non-surgical) management, particularly when the cranial deformity is subtle and unaccompanied by raised intracranial pressure [8].

A variety of surgical techniques, ranging in invasiveness, are employed to release fused sutures, correct craniofacial asymmetry, and facilitate normal brain growth. These include minimally invasive endoscopic suturectomy as well as more extensive open cranial vault remodelling and reconstructive procedures [9].

The primary goal of surgical correction in craniosynostosis is to relieve any potential restriction on brain growth and to restore normal skull shape. However, the timing of surgery has emerged as a key determinant of cognitive and functional outcomes. Studies have consistently shown that early intervention, particularly before 6 months of age, is associated with improved neurocognitive development and lower risk of long-term impairment [10,11].

The rationale is based on the concept of neuroplasticity during infancy, where the brain has a greater capacity to reorganize and compensate when structural correction is achieved early [12].

In addition to surgical timing, the number and type of affected sutures have been linked to outcome variability. Patients with multi-suture or complex craniosynostosis often have poorer developmental trajectories, likely due to the compounded effect of restricted cerebral expansion and associated anomalies [13,14].

Conversely, isolated single-suture craniosynostosis, particularly of the sagittal or unicoronal types, tends to be associated with better cognitive outcomes, especially when treated early [15,16].

To better understand the interplay between these factors, our study evaluated a cohort of children with non-syndromic craniosynostosis, assessing their cognitive abilities using the Stanford-Binet Intelligence Scales, Fifth Edition (SB5), and psychomotor function using the Assessment and Programming of Psychomotor Skills (APPS). We investigated how age at surgery, type and number of sutures involved, and surgical approach influenced developmental outcomes, providing further evidence to inform surgical decision-making and long-term management.

Aim of the work
This work aims to investigate the potential factors influencing functional outcomes in patients undergoing craniosynostosis surgery. It seeks to determine whether there is a relationship between the age at the time of surgical intervention and the postoperative functional outcome. Additionally, it explores whether the type of surgical intervention performed correlates with the functional outcome. Finally, the study examines whether the number and type of affected cranial sutures have an impact on the functional outcome. By addressing these questions, this work seeks to provide a clearer understanding of prognostic factors that may guide surgical decision-making and improve patient outcomes.

Patients and Methods
This retrospective analytical study was conducted on patients diagnosed with craniosynostosis who underwent surgical intervention at the Neurosurgery Department of El Sahel Teaching Hospital, Cairo, between July 2009 and July 2020. During this period, 79 patients were operated upon. Following a thorough

review of medical records, 58 cases met the inclusion criteria and were enrolled in the study. Inclusion was limited to patients diagnosed with primary non-syndromic craniosynostosis, while 21 cases with secondary craniosynostosis owing to cerebral trophy, hydrocephalus, and/or syndromic cases were excluded.

The final cohort consisted of 32 males and 26 females. Surgical techniques varied based on patient age, suture involvement, and deformity severity, and included endoscopic-assisted suturectomy, open corrective surgery, or a combination of both approaches. The age at the time of surgery ranged from 2 to 36 months.

To assess long-term outcomes, each patient underwent a standardized functional evaluation five years postoperatively. Evaluations were conducted by a certified Applied Behavior Analysis (ABA) therapist and focused on assessing the integrity of higher cortical functions, locomotor abilities, and learning performance. Special attention was given to analyzing the relationship between the age at the time of surgery, the type of surgery performed, the number and type of affected sutures, and the functional outcome.

The following functional tests are used:

1- The Stanford-Binet Intelligence Scales, Fifth Edition (SB5) was used to assess the cognitive and intellectual functioning of the patients. This standardized test evaluates a range of mental and cognitive abilities, including intelligence quotient (IQ), mental age, memory, concentration, visual processing, and reasoning. It provides a comprehensive profile of cognitive strengths and weaknesses in relation to age-appropriate norms. IQ scores obtained from the SB5 are classified as follows: Superior: IQ 120–129, High Average: IQ 110–119. Average: IQ 90–109, Low Average: IQ 80–89, Borderline Impaired or Delayed: IQ 70–79, Mildly Impaired or Delayed: IQ 55–69, Moderately Impaired or Delayed: IQ 40–54, Severely Impaired or Delayed: IQ ≤ 39. This classification system allowed for objective comparison of the cognitive outcomes of the studied patients in relation to age-matched normative data [17].

2- The Assessment and Programming of psychomotor Skills (APPS) test was utilized to evaluate the psychomotor processing abilities of the patients. This assessment covers three Key domains: Body, spatial, and temporal (time) awareness. Each domain is evaluated and scored using a numerical rating scale ranging from 0 to 4, with the total score reflecting the child's overall level of psychomotor independence and functionality. The scoring system is interpreted as follows: 1-Score 0: Very weak in all practical skills; the child is entirely dependent and unable to care for themselves, requiring complete assistance. 2-Score 1: Weak in all practical skills; the child is partially able to care for themselves but still requires considerable support. 3-Score 2: Weak in all practical skills; however, the child can care for themselves independently without external help. 4-Score 3: Intermediate performance in practical skills; the child can independently care for themselves. 5-Score 4: Fully functional in all practical skills; the child demonstrates complete independence in daily activities. The APPS test provided a structured measure of each child's psychomotor development and independence in daily life activities, supporting the evaluation of long-term functional outcomes [18].

Statistical analysis

The collected data were analyzed using the Statistical Package for the Social Sciences (SPSS), version 25.0 (IBM Corp., Chicago, USA). The distribution of variables was assessed using the Kolmogorov-Smirnov test to determine normality. For normally distributed data, comparisons were made using the independent samples t-test and the chi-square test where appropriate. For non-normally distributed (heterogeneous) variables, non-parametric statistical methods were applied. These included the Kruskal-Wallis test for comparing more than two groups and the Mann-Whitney U test for comparing two independent groups. A p-value of less than 0.05 was considered statistically significant.

Results

The mean age at the time of surgery for the patients included in our study was 6.79 ± 4.9 months, ranging from 2 months to 36 months. Thirty-two patients were males (55.2%) and twenty-six were females (44.8%). Regarding the type of involved sutures, bicoronal suture involvement was the most common form among patients enrolled in the study (43.1%), followed by sagittal suture involvement (12.1%), metopic-bicoronal (10.3%), unicoronal (10.3%), and small percentages of other forms of suture involvement were also included in the study (Table 1). Regarding the number of sutures involved, 48.3% of the patients had two sutures, 32.8% had only one affected suture, 13.8% had three sutures involved, and 5.2% had pansynostosis (Table 2). Forty-six patients (79.3%) underwent suturectomy, while 12 patients (20.3%) underwent reconstructive surgery (Table 3). The overall mean SB5 for all the patients enrolled in the study was 90.34 ± 9.62, while the mean APPS were 3.81 ± 0.48.

Table 1: Types of involved sutures.

Involved Sutures		
	Frequency	Percent
Bicoronal	25 patients	43.1
Bicoronal, sagittal	1 patient	1.7
Bilambdoid	1 patient	1.7
Metopic	4 patients	6.9
Metopic , bicoronal	6 patients	10.3
Metopic , sagittal	1 patient	1.7
Pansynostosis	3 patients	5.2
Sagittal	7 patients	12.1
Unicoronal	6 patients	10.3
Unicoronal, Metopic	1 patient	1.7
Unicoronal, unilambdoid	1 patient	1.7
Unilambdoid	2 patients	3.4
Total	58 patients	100.0

Table 2: Number of involved sutures in the studied group.

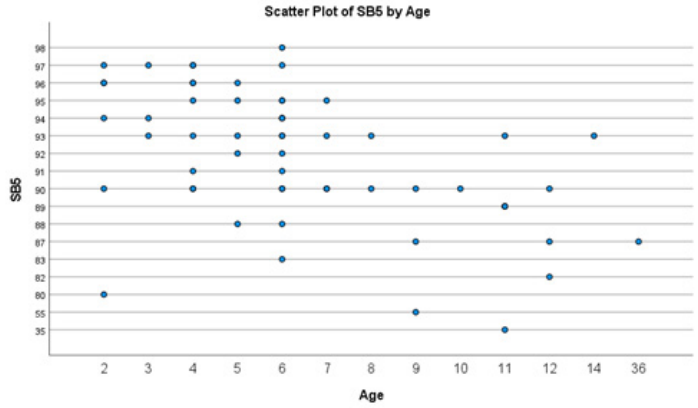
Number of involved Sutures			
		Frequency	Percent
No. of Sutures Involved	1	19 patients	32.8
	2	28 patients	48.3
	3	8 patients	13.8
	6 (Pansynostosis)	3 patients	5.2
	Total	58 patients	100.0

Table 3: Type of surgical intervention.

Type Of Surgery			
		Frequency	Percent
Surgery	Reconstructive	12	20.7
	Suturectomy	46	79.3
	Total	58	100.0

Correlation between age at surgery and functional Outcome

It has been found that there was a strong negative correlation between age and SB5 (Correlation Coefficient: -0.521), meaning that the younger the age at time of surgery, the better the functional outcome as measured by SB5, and this correlation was statistically significant (P <0.001). On the other hand, there was also a weak negative correlation between age and functional outcome as measured by APPS (Correlation Coefficient: -0.214). However, this correlation was not statistically significant (P 0.107) (Graph 1).



Graph 1: Scatter plot of SB5 and age.

Patients were divided into three subgroups based on their age at the time of surgery. The 1st group was: age ≤ 6 months, the 2nd group was: age >6-<12 months, and the 3rd group was: age ≥ 12 months (Table 4). There was a statistically significant difference between the three groups in comparing functional outcome as measured by SB5 (P<0.001). It has been found that patients who had surgeries at age ≤6 months were associated with higher SB5 values either when compared with the age group >6-<12 months (P<0.001) or the age group ≥12 months (P=0.007). Data analysis also showed that there was no statistically significant difference between the age group >6-<12 months and the age group ≥12 months (P 0.445) (Table 5). There was no statistically significant difference between different age groups regarding APPS (P 0.278). Nevertheless, APPS scores were slightly higher in patients aged ≤6 months.

Table 4: Age groups.

Age Groups			
		Frequency	Percent
Groups	≤6 months	38	65.5
	>6-<12 months	15	25.9
	≥12 months	5	8.6
	Total	58	100.0

Table 5: SB5 in Different Age Groups.

SB5 in Different Age Groups				
	N	Mean	Std. Deviation	P Value
≤6 months	38	92.97	3.838	P < 0.001
>6-<12 months	15	84.53	16.617	
≥12 months	5	87.80	4.087	
Total	58	90.34	9.624	

Effect of Number of Affected Sutures on Functional Outcome

In the context of the number of sutures involved, there was a negative correlation between the number of affected sutures and functional outcome as measured by SB5 (Correlation Coefficient: -0.46). Fewer sutures involved were associated with better outcomes in SB5, and this correlation was statistically significant (P <0.001) (Table 6). Similarly, the number of affected sutures was associated with better APPS scores and this effect was of statistical significance (P <0.001) (Table 7). The number of sutures involved also affected the functional outcome as measured by SB5 and APPS. The fewer the affected sutures, the higher the SB5 score (P 0.004). Cases with 1 or 2 sutures affected had statistically significantly better outcomes than cases with>2 sutures involved.

Table 6: No. of sutures affected vs SB5.

	No. of sutures affected vs SB5			
No. of involved sutures	N	Mean	Std. Deviation	P Value
1	19	93.53	3.306	0.004
2	28	91.11	7.583	
3	8	88.00	3.780	
6	3	69.33	30.436	
Total	58	90.34	9.624	

Table 7: No. of sutures affected vs APPS.

No. of sutures affected vs APPS				
No. of sutures affected	N	Mean	Std. Deviation	P Value
1	19	4.00	.000	<0.001
2	28	3.86	.448	
3	8	3.50	.535	
6	3	3.00	1.000	
Total	58	3.81	.476	

Effect of the type of affected sutures on functional outcome

As regards the effect of the type of involved sutures on functional outcome, there was a difference of statistical significance between different groups (P 0.038). Unilambdoid suture and metopic suture

groups were associated with the highest SB5 values, whereas pansynostosis and metopic-bicoronal groups had the lowest SB5 values (Table 8). Similarly, there is a statistically significant difference between the types of affected suture groups as regards APPS (P=0.027). Data Analysis showed that the type of sutures involved affects functional outcomes, either SB5 or APPS.

Table 8: SB5 vs Type of Suture Involved.

SB5 vs Type of Suture Involved				
	N	Mean	Std. Deviation	P Value
Bicoronal	25	91.16	8.040	
Bicoronal, sagittal	1	93.00	.	
Bilambdoid	1	91.00	.	
Metopic	4	95.75	.957	
Metopic, bicoronal	6	86.83	3.545	
Metopic, sagittal	1	90.00	.	
Pansynostosis	3	69.33	30.436	.038
Sagittal	7	93.14	2.410	
Unicoronal	6	91.67	4.546	
Unicoronal, metopic	1	90.00	.	
Unicoronal, unilambdoid	1	91.00	.	
Unilambdoid	2	96.00	1.414	
Total	58	90.34	9.624	

Effect of type of surgery on functional outcome

Eventually, it has been noticed that the type of surgery also does influence functional outcome. Cases that underwent suturectomy were associated with better outcomes, and this difference was statistically significant. Suturectomy was associated with better SB5 values (P <0.001) and APPS values (P 0.04).

Discussion

Despite expert consensus on the value of early intervention, there remains a paucity of robust evidence directly linking early surgical management to long-term functional outcomes. This study helps bridge that gap by providing evidence on the actual relationship between the timing of intervention and sustained neurocognitive and psychomotor improvement. Moreover, the research highlights the critical role of surgical variables, including the type of procedure performed and the type and number of sutures affected, in shaping functional outcomes. By addressing these dimensions, the study moves beyond cosmetic or morphological considerations and emphasizes the functional and developmental implications of surgical decision-making. Importantly, the use of standardized psychomotor and Stanford assessments provides objective and reliable measures of patient progress, strengthening the validity of the findings. The results, therefore, not only add depth to the current body of literature but also offer practical insights that may inform clinical guidelines, optimize surgical planning, and improve prognostic counselling for families.

The findings of this study support the growing consensus that early surgical intervention in cases of non-syndromic craniosynostosis significantly improves long-term neurodevelopmental outcomes. Our data revealed a statistically significant inverse relationship between age at time of surgery and cognitive outcome as measured by the Stanford-Binet Intelligence Scales, Fifth Edition (SB5). Children who underwent surgery at or before 6 months of age demonstrated significantly higher SB5 scores than those operated on at older ages. This aligns with the existing literature, indicating that surgical correction before 6 months allows more effective brain growth and minimizes the risk of elevated intracranial pressure and restricted cortical development [10,15].

Although the association between age and psychomotor outcomes as measured by the APPS was weaker and not statistically significant, a trend toward improved psychomotor function with earlier intervention was still evident. This may reflect the more plastic and adaptable nature of the infant brain, particularly within the first year of life [12]. Other studies similarly report improved language and motor development in infants treated earlier [11].

The type and number of sutures involved also had a significant impact on functional outcomes. Patients with fewer sutures involved had better SB5 and APPS scores, consistent with previous studies showing that multi-suture involvement is associated with greater structural and possibly functional brain compromise [13].

Notably, pansynostosis and metopic-bicoronal synostosis were associated with the lowest SB5 outcomes, likely due to the extensive restriction they impose on cranial and cerebral expansion. Conversely, children with isolated unicoronal or metopic synostosis tended to have better outcomes, reinforcing prior findings that single-suture craniosynostosis, particularly when corrected early, carries a more favourable prognosis [16].

Furthermore, the type of surgery performed emerged as a determinant of functional outcomes. Our data indicate that patients who underwent suturectomy alone had significantly better SB5 and APPS scores compared to those who underwent reconstructive procedures. This finding is in line with recent literature suggesting that minimally invasive techniques such as endoscopic-assisted suturectomy, particularly when performed early, reduce surgical morbidity and better support neurodevelopment [19,20]. However, this must be interpreted cautiously, as the need for more extensive reconstructive surgery may correlate with more complex presentations or delayed diagnosis. Overall, our findings support the strategy of early detection and early surgical correction, particularly in patients with single-suture involvement. Long-term neurodevelopmental follow-up and functional assessment, including intelligence and psychomotor scales, remain critical in guiding postoperative care and parental counselling.

Limitations

Some limitations should be acknowledged. As the retrospective nature of the study may introduce selection bias. Also, the sample size, while reasonable, limits subgroup comparisons, especially among rarer types of synostosis. Furthermore, socioeconomic and educational factors, which may influence developmental outcomes, were not controlled for in this analysis.

Conclusion

Early surgical intervention, fewer affected sutures, and the use of suturectomy were all associated with better long-term cognitive and psychomotor outcomes in children with non-syndromic craniosynostosis. Adopting the utilization of Stanford-Binet Intelligence Scales, and the Assessment and Programming of psychomotor Skills to evaluate cognitive and psychomotor outcomes of such cases periodically before and after the procedure on regular visits. These findings highlight the importance of early detection, individualized surgical planning, and the consideration of less invasive techniques when appropriate. Future prospective, multicentre studies with larger cohorts and long-term neurodevelopmental follow-up are recommended to further validate and expand upon these findings.

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