Original Article

Assessing Long-Term Neurodevelopment among Children with Non-Syndromic Single Suture Craniosynostosis

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ABSTRACT

Background: Single suture craniosynostosis (SSC) is a disorder, affecting brain growth. Reviewing literature reveals controversialists of papers in this field.

Methods: This prospective study was conducted from 2014 to 2016. All the individuals, aged 2 to 16 years, whose medical records files were complete, with SSC from 1999 to 2013 were included. All patients had undergone cranial vault remodeling at Mofid Hospital, Tehran, Iran. Wechsler questionnaires, WPPSI-III and WISC-IV, were completed for each child based on his/her age.

Results: Seventy children were included, with the mean age of 6.7 (±2.9) years. Forty-six (65.7%) children were boys while 24 (34.3%) were girls. Mean FSIQ for all of children was 95.5 (±13.2). Mean verbal IQ, performance IQ, verbal comprehension, perceptual reasoning, processing speed, and working memory are 93.4 (±14.1), 96.1 (±13.3), 97.5 (±13.9), 102.2 (±12.5), 94.5 (±9.8), and 97.5 (±12.9), respectively. There was statistically significant difference between FSIQ of children with SSC and that of unaffected children (P-value<0.05). There was significant difference between verbal IQ of children with SSC and that of unaffected ones (P-value< 0.007). There was significant difference between in processing speed between affected children and unaffected children (P-value<0.012).

Conclusion: Children, aged 2 to 6 years, with SSC had a significantly lower Verbal IQ, and children, aged 6 to 16 years, with SSC had a significantly lower processing speed than their healthy counterparts. Though FSIQ of children with SSC falls within normal range, it is a little lower than healthy peers.

Keywords: IQ; Single suture craniosynostosis; Neurodevelopment, WISC, WPPSI, Wechsler.

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INTRODUCTION

Craniosynostosis is a pathologic condition characterized by premature fusion of one or more cranial sutures, leading to abnormal shape of skull. Single suture craniosynostosis (SSC) is defined when only one suture—either sagittal, metopic, right or left coronal, or right or left lambdoid— is prematurely fused [1, 2]. Craniosynostosis is divided into syndromic or nonsyndromic. Syndromic craniosynostosis—accompanied by extracranial anomalies—is less frequent than nonsyndromic craniosynostosis. Moreover, in contrast to nonsyndromic

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craniosynostosis, syndromic craniosynostosis most often involves multiple sutures of cranial [3-5].

A number of patients with craniosynostosis may develop, either short or long term, some complications. Increased intracranial pressure (IICP) which commonly affects patients with multiple suture craniosynostosis is a serious complication. However, IICP may lead to papilledema and optic nerve atrophy causing visual disruption. Although hydrocephalus is an uncommon complication in these patients, patients with syndromic craniosynostosis are more prone to develop it. Neurodevelopmental delay is a major concern in such patients. Several craniofacial syndromes are associated with both neurodevelopmental and intellectual delay, but, up to now, the definite etiology is not obvious [4, 6-10].

Reviewing literature reveals that reports on different aspects of intellectual functioning in these patients are controversial. Many studies have demonstrated that such patients may suffer from lower development/function than normal counterparts, but some studies have showed that these patients have normal function as healthy ones [11-13].

Herein, we conducted a prospective study to assess the intellectual functioning among patients with craniosynostosis in several age groups in comparison to normal counterparts.

MATERIALS AND METHODS

This prospective study was conducted from 2014 to 2016. All the individuals, aged 2 to 16 years, whose medical records files were complete, with SSC from 1999 to 2013 were included. All patients had undergone cranial vault remodeling at Mofid Hospital, Tehran, Iran. Those patients agreed to participate in the study were interviewed by a psychologist. Three different questionnaires, based on their age group, were used.

This study was approved by ethical committee of Shahid Beheshti University of Medical Sciences (SBMU; Ethical Registration Number: SBMU.REC.1392.258).

Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III)

The Wechsler Preschool and Primary Scale of Intelligence, consisting of 14 subtests, was designated by Wechsler. Subsets are one of three following types: core, supplemental, or optional. The core subtests are used to calculate the Verbal, Performance, and Full Scale IQ. The supplemental subtests provide additional information about cognitive abilities or can be used as replacement for inappropriate subtests. The optional subtests are those which are providing additional information about cognitive functioning but cannot be used as replacements for core subtests [14].

Wechsler Intelligence Scale for Children–Fourth Edition (WISC-IV)

The WISC-IV is a test assessing intellectual ability, only individuals aged 6 to 16 years. This test contains 15 subtests while each one is allocated to either the Verbal Comprehension (VC), Perceptual Reasoning (PR), Working Memory (WM), or Processing Speed (PS) subscales. Each subscale has a standardized mean and SD of 100 and 15, respectively. The Full Scale IQ (FSIQ) is consists of 10 core sub tests: 3 VC (Vocabulary, Similarities, and Comprehension), 3 PR (Block Design, Picture Concepts, and Matrix Reasoning), 2 WM (Digit Span and Letter-Number Sequencing), and 2 PS (Coding and Symbol Search). This scale, including the FSIQ, four indices, and subsets, has high reliability and validity [15-17].

Data Analysis

All statistical analyses were performed using statistical package for social science (SPSS 16, Chicago, IL, USA). Variables are expressed as number (%) or mean (±SD). One sample *t*-test was used in calculating *P*-value for FSIQ, verbal IQ, performance IQ, verbal comprehension, perception reasoning, processing speed, and working memory. A *P* value less than 0.05 was considered statistically significant (*P*-value<.05).

RESULTS

A total of 130 patients were informed, but only 70 (53.8%) patients agreed to participate in the study. Forty-six (65.7%) children were boys while 24 (34.3%) were girls, with the mean age of 6.7 (±2.9) years (Table 1). Mean FSIQ for all of children was 95.5 (±13.2), ranging from 65 to 122. Thirty-one (44.2%) of patients had an IQ from 86 to 100 (Table 2). As Table 3 outlines, mean verbal IQ, performance IQ, verbal comprehension, perceptual reasoning, processing speed, and working memory are 93.4 (±14.1), 96.1 (±13.3), 97.5 (±13.9), 102.2 (±12.5), 94.5 (±9.8), and 97.5 (±12.9), respectively. There was statistically significant difference between FSIQ of children with

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Table 1: Demographics of children based on each relevant questionnaire.

Variable		WPPSI-III 2-6 years	WISC-IV 6 to 16 years
Number of Children (%)		42 (60%)	28 (40%)
Mean age (±SD)		3.7 (±0.79)	9.6 (±2.4)
Sex	Female	11 (26.1%)	13 (46.4%)
	Male	31 (73.8%)	15 (53.6%)

Table 2: Number of children in each FSIQ group and mean of IQ in each one.

Variable	Full Scale IQ				
	<70	71-85	86-100	101-115	>115
Number of Children (%)	2 (2.8%)	13 (18.5%)	31 (44.2%)	21 (30%)	3 (4.2%)
Mean FSIQ (±SD)	65.5 (0.7)	77.8 (3.15)	94.2 (±5.1)	107.7 (±3.9)	120 (±3.4)

Table 3: Outcomes of Wechsler's questionnaires.

Variable		Number of children	Mean (±SD)	Median	Minimum	Maximum	*P*-value
FSIQ		70 (100%)	95.5 (±13.2)	95	65	122	0.006
WPPSI-III	Verbal IQ	42 (60%)	93.4 (±14.1)	95	66	122	0.007
	Performance IQ		96.3 (±13.3)	96	67	119	0.09
WISC-IV	Verbal comprehension	28 (40%)	97.5 (±13.9)	99	75	130	0.39
	Perception reasoning		102.2 (±12.5)	104	77	127	0.38
	Processing Speed		94.5 (±9.8)	98	78	117	0.012
	Working Memory		97.7 (±12.9)	96	76	120	0.402

SSC and that of unaffected children (*P*-value: 0.006, t: -2.8, df: 69). There was not statistically significant difference between performance IQ of children with SSC and that of normal children (*P*-value: 0.09, t: -1.7, df: 41). There was significant difference between verbal IQ of children with SSC and that of unaffected ones (*P*-value: 0.007, t: -2.8, df: 41). There was no significant difference between working memory of affected children and that of normal ones (*P*-value: 0.402, t: -0.85, df: 27). There was significant difference between in processing speed between affected children and unaffected children (*P*-value: 0.012, t: -2.7, df: 27). There was no significant difference in perceptual reasoning between children with SSC and normal children (*P*-value: 0.38, t.88, df: 27). There was no significant difference in verbal comprehension between affected children and unaffected children (*P*-value: 0.39, t: -0.86, df:27).

DISCUSSION

In contrast to many craniofacial disorders, craniosynostosis is a common congenital disorder of craniofacial region, with an incidence of 1 in 2000 to 2500 births [5, 18, 19]. Majority of cases, approximately 85%, occur without any genetic background, most probably due to a spontaneous mutation [20, 21].The diagnosis is often made within the first months

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of life, and the mainstay of treatment is surgical approach in which the fused suture/sutures are released to reshape the calvarium, allowing brain growth and normal neurodevelopment. Although the precise time of surgery is debated, craniotomy is performed for all patients in the first year of life [22]. However, based on some studies, these children may show some neurodevelopment delays. In here, FSIQ of children with SSC was around 95, and there was statically significant difference between FSIQ of patients with craniosynostosis and unaffected children (*P*-value<0.05). FSIQ of children with SSC is significantly lower than normal counterparts, but their FSIQ falls within normal range. Children, aged 2 to 6 years, with SSC had significantly lower verbal IQ, some 93, than unaffected peers (*P*-value<0.05). Moreover, processing speed, approximately 94, of children, aged 6 to 16 years, with SSC was significantly lower than normal ones (*P*-value<0.05). Yet, in performance IQ, verbal comprehension, and working memory the difference was not statistically significant (*P*-value>0.05). Although children with SSC had a little higher perception reasoning, the difference was not statistically significant.

Chieffo et al evaluated 65 children who had undergone surgery for sagittal or unicoronal craniosynostosis concluded that such children will manifest lower than average 6 cognitive level [23]. Mendonca et al demonstrated that 30% of patients with metopic synostosis had speech and language delays, and delays were not associated with severity of disorder [24]. Shipster et al evaluated 76 children with isolated sagittal synostosis and concluded that such children are at increased risk developing speech and language delays [25]. Magge et al retrospectively reviewed 16 patients with sagittal synostosis and found out that their IQ was in normal range but learning disabilities were significantly prominent within them [26]. Boltshauser et al prospectively assessed 30 children with isolated sagittal synostosis, and concluded that their school performance, behavior and quality of life were reassuring[27]. Kelleher et al assessed 63 children with trigonocephaly, concluded that they had high frequency of developmental, educational, and behavioral problems [28]. Wallace ER et al evaluated 179 children with SSC and concluded that such children have deficits in manual dexterity but their visual processing are similar to that of normal ones [22]. The cognitive, behavioral, and psychological outcomes of children with metopic synostosis were worse than their normal counterparts [29]. There was no significant difference between healthy children and children with SSC in language, memory, and learning tasks in early elementary school ages [30]. FSIQ of children in a study was around 96 which was similar to normal children, and, in addition, VIQ was greater than PIQ [31]. Collett et al in a prospective study evaluated 179 school-aged children, but their study outcomes were variable to definitely conclude [32]. In another study it is indicated that there is no definitive relation between size of brain and intelligence, so this is the reason why FSIQ of children is not far lower than healthy counterparts [21].

CONCLUSION

Children, aged 2 to 6 years, with SSC had a significantly lower Verbal IQ, and children, aged 6 to 16 years, with SSC had a significantly lower processing speed than their healthy counterparts. However, children with SSC had a higher perception reasoning than healthy peers. Although FSIQ of children with SSC falls within normal range, it is a little lower than healthy peers.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interests.

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