RESEARCH BRIEF

Diagnostic Accuracy of Ultrasonic Examination in Suspected Craniosynostosis Among Infants

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The current study was performed to assess the diagnostic accuracy of ultrasound compared to CT scan as a gold standard in the diagnosis of craniosynostosis. 44 infants (17 girls) under 1 year old, clinically suspected to have craniosynostosis, were first sonographically examined by a pediatric radiologist and were later referred to another blinded pediatric radiologist to examine CT scan with 3D reconstructed images of skull. Sensitivity, specificity, positive and negative predictive values of ultrasound *versus* CT scan were 96.9%, 100%, 100%, and 92.3%, respectively. The high specificity of ultrasound helps to correctly rule out craniosynostosis in clinically suspected cases and thus, can prevent unnecessary exposure of healthy infants to CT scan ionizing radiation.

Key words: *Craniosynostosis, Diagnosis, Infants, Ultrasonography.*

Craniosynostosis is defined as premature fusion of cranial sutures. It occurs in 4-6 of 10,000 live births [1]. Primary craniosynostosis is mostly idiopathic and is due to premature fusion of cranial sutures, while the secondary type is the result of brain defects, including brain microcephaly or atrophy which leads to premature fusion of the sutures [1]. In simple craniosynostosis only one suture is involved and in compound type, more than one suture are involved [2-4].

Craniosynostosis is traditionally diagnosed by imaging modalities and in-time diagnosis of primary cases is pivotal in the success of surgical treatment. Standard radiographs are the first step in the evaluation of suspected cases [5,6]. Presently, CT scan is considered as an alternative to standard radiography [1]. CT scan can differentiate primary from secondary cases of craniosynostosis by providing adequate information about brain parenchyma. However, a major disadvantage is the high radioactive dose [7,8], in addition to the cost and availability issues.

Ultrasound is a non-invasive, available, low-cost and safe modality, and is a plausible alternative to CT scan in the diagnosis of craniosynostosis [9]. The diagnostic accuracy of ultrasound is not established. It may even be used for prenatal diagnosis of craniosynostosis [11]. In cases which fontanels are open, ultrasound can reveal reliable information about the brain structures also. The current study has been performed to investigate the diagnostic accuracy of ultrasound in detecting premature fusion of sutures among infants under one year of age.

METHODS

Symptomatic infants from urban areas of Tehran were referred to Tehran Children's Medical Center, where they were examined by a pediatric neurologist or a pediatric neurosurgeon. The inclusion criteria was any suspected cases who had small head circumference or had a head skull deformity. Informed consent was obtained from parents before inclusion of infants in the study. From June 2007 to September 2008, 44 infants under 1 year of age, clinically suspected with craniosynostosis, were included in the study and were examined first by ultrasound and then by CT scan. In case the infant was restless, chloral hydrate was administered for sedation. We did not use oral sedation for ultrasound exams but it was administered to 10 children for CT scan. Radiologists who interpreted the CT scan were blinded to the diagnosis made on ultrasound. CT scan was performed hellically with a 16 slice GE apparatus with a thickness of 5 mm at an interval of 4 mm, and was reconstructed with a thickness of 1.25 mm and the interval of 1 mm before 3D-skull reconstruction. The routine condition is KV 120 and mA 45 to 80.

All sonographic examinations were performed using an Ultrasonix machine with a 14 MHz linear probe, in a near field focus and with a depth of 2 cm. The probe was placed vertically on each suture and the whole length of sutures were evaluated. The cranial suture was

considered normal in case a hypoechoic or beveled gap between 2 hyperechoic bones was noticed. The absence of hypoechoic gap, beveled appearance, or the presence of ridging or bridging along the bone was considered abnormal. Also narrow sutures, the width of which was less than 0.5 mm, were considered as abnormal (0.5 mm is the narrowest distance measurable by examiner). The diagnostic criteria used have been detailed previously [12]. Data were analyzed in SPSS version 16.0 (Chicago, IL) and the sensitivity, specificity, positive and negative predictive values of ultrasound were calculated.

RESULTS

Forty four infants (17 girls) were included. With mean age of 5 months and 23 days (SD 3 months and 13 days), (Range, 18 days - 12 months). The mean head circumference was 41.1(SD 3.33) cm, (range, 34-47.5 cm). Only 5 infants (11.4%), all of whom were boys, had familial history of craniosynostosis. Delivery was normal in 16 infants (36.4%) and cesarean section in 28 infants (63.6%). There was no difference in mean age and mean head circumference between sexes, neither any difference in mean head circumference between infants born normally and those by cesarean section.

Sonographic findings: Thirteen infants (29.5%) were recognized as healthy and 31 infants (70.5%) were diagnosed as cases of craniosynostosis. There was no difference in female-to-male ratio, the mean age, the mean head circumference, and percentage of cesarean section among healthy and unhealthy infants. Craniosynostosis was primary in 29 infants (93.5%) and secondary in 2 infants (6.5%). Craniosynostosis was simple in 27 infants (87.1%) and compound in 4 infants (12.9%). The most prevalent sutures involved, ordered from high to low included: metopic, sagital, unilateral and bilateral coronal, and bilateral lambdoid. Cranial deformities ordered from high to low prevalence included: trigonocephaly, scaphocephaly, brachycephaly, anterior plagiocephaly, and posterior plagiocephaly. The brain was abnormal in 2 infants (6.5%).

In the current study, the diagnosis of 43 infants in ultrasound was completely compatible with CT scan. Only one patient diagnosed in CT scan was missed in ultrasound. The patient was a boy with 7 and half months of age, with positive family history, and born by cesarean section. The boy had primary compound craniosynostosis detected in CT scan, involving both sagital and metopic sutures who presented with scaphocephaly. In CT scan, bridging was noticed along 4 cm of the suture. The brain was reported to be normal.

The sensitivity, specificity, positive and negative predictive values of ultrasound versus CT scan were 96.9%, 100%, 100%, and 92.3%, respectively. There was no significant difference in diagnostic accuracy of ultrasound between girls and boys, and between infants under 6 months and infants older than 6 months **(*Table* I)**.

DISCUSSION

The higher prevalence of simple craniosynostosis than compound type, and primary than secondary type is concordant with previous studies [1,2]. It may be a referral bias, as primary cases of craniosynostosis are more frequently referred to Children's Medical Center than secondary cases, because primary cases can be surgically treated. The relative frequency of sutures involved and of various types of cranial deformities was not concordant with previous studies [13]. One possible justification is that scaphocephaly is not considered as a skull deformity among general public, and that's why most referred infants present with trigonocephaly.

We emphasize the high negative predictive value of

TABLE I DIAGNOSTIC CHARACTERISTICS OF ULTRASOUND COMPARED TO CT SCAN

	Gender		*Age*		
	Female (n=17)	*Male (n=27)*	*< 6 mo (n=26)*	*>6 mo (n=18)*	*Total*
Sensitivity (95% CI)	100% (97.0%-100%)	95.2% (90.5%-99.9%)	100% (97.8%-100%)	90.9% (81.8%-100%)	96.9% (93.8%-100%)
Specificity (95% CI)	100% (95.9%-100%)	100% (95.9%-100%)	100% (96.3%-100%)	100% (95.6%-100%)	100% (97.1%-100%)
Positive Predictive Value (95% CI)	100% (97.0-100%)	100% (97.8%-100%)	100% (97.8%-100%)	100% (96.9%-100%)	100% (98.2%-100%)
Negative Predictive Value (95% CI)	100% (95.9%-100%)	85.7% (71.4%-100%)	100% (96.3%-100%)	83.3% (66.6%-100%)	92.3% (84.7%-99.9%)

CI: Confidence Interval.

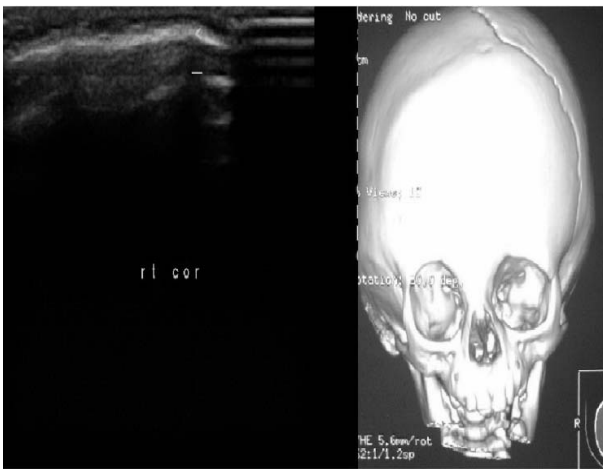

FIG.1 *(a) Sonographic image and (b) CT scan image of a one-month old female infant with right coronal synostosis and plagiocephaly.*

ultrasound compared to CT scan. Ultrasound can be suggested as the preferred screening method for craniosynostosis as many patients will be thus spared from unnecessary exposure to ionizing radiation, as also suggested earlier [14]. The accuracy reported in previous studies is even higher that the current study [10,12,14]. Moreover, ultrasound is capable of diagnosing prenatal craniosynostosis [11]. However, it should be noted that ultrasound is operator-dependent and radiologists should be specifically trained to detect craniosynostosis by using ultrasound [15].

The overall accuracy of ultrasound justifies its applicability in the diagnosis of craniosynostosis. Based on the results of this study, we can conclude that ultrasound can be a low-cost and accurate alternative to CT scan, especially the preferred screening method in infants clinically suspected to primary simple or complex craniosynostosis.

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