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Evaluation of mortality scores in patients with adult congenital heart defects

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ABSTRACT

Objectives: Currently, there is no approved risk stratification for adult congenital heart surgery; accordingly, this study aimed to evaluate risk stratification for congenital heart surgery in the pediatric age in terms of its prognostic value in adult patients as well as the effectiveness of the newly developed Adult Congenital Heart Surgery (ACHS) score in this patient group.

Patients and methods: A total of 205 patients (115 males, 90 females; mean age: 25.0±11.4 years; range, 18 to 65 years) operated on due to congenital heart disease between January 1, 2011, and August 1, 2019, were studied retrospectively. Aristotle Basic Complexity (ABC) score, Society of Thoracic Surgery European Association for Cardiothoracic Surgery (STAT) score, and ACHS score were evaluated. Receiver operating characteristic (ROC) curves were created to evaluate the ability of scoring systems to predict mortality.

Results: The mortality rate was 4.4% (n=9). For mortality, areas under the ROC curve were 0.89, 0.89, and 0.70 for ABC, STAT, and ACHS scores, respectively. The mean ACHS score was 0.42±0.34. The cut-off point of the ACHS score was identified as 0.7 and above. The specificity of the cut-off value of 0.7 regarding the ACHS score was 94.39%. Adult Congenital Heart Surgery scores were found to be statistically high in patients with mortality ($p=0.037$; $p<0.05$).

Conclusion: Adult Congenital Heart Surgery scores had higher specificity in determining mortality in cases with an ACHS score of 0.7 and above. The ACHS score could also be used to determine the expected mortality rate, similar to the ABC and STAT scores.

Keywords: Adult congenital heart surgery score; aristotle basic complexity score; mortality; society of thoracic surgery european association for cardiothoracic surgery score.

In congenital heart disease, Aristotle Basic Complexity (ABC) and Society of Thoracic Surgery European Association for Cardiothoracic Surgery (STAT) scores are frequently used to determine mortality. In grown-up congenital heart (GUCH) patients, surgery is complex due to residual lesions, complications, or sequelae after previous palliative surgery or complete correction.[1]

Previous studies reported that GUCH patients had several risk factors for morbidity and mortality after surgery. Mortality rates detected in GUCH patients are low; however, our knowledge about this growing population of patients is still limited.[2] This study aimed to determine the effectiveness of the Adult Congenital Heart Surgery (ACHS) score for early mortality in primary surgery and reoperations and compare it with ABC and STAT scores.

PATIENTS AND METHODS

In this retrospective study, 205 patients (115 males, 90 females; mean age: 25.0±11.4 years; range, 18 to 65 years) operated on due to congenital heart disease at the Kartal Koşuyolu Yüksek Ihtisas Training and Research Hospital between January 1, 2011, and August 1, 2019, were included. The patients were divided into 27 subgroups according to the type of operation. In our study, ABC and STAT mortality scores of each patient were calculated, and an ACHS score was given according to the procedures performed on the patients based on the studies conducted by Fuller et al.[1] in 2015. Patients who underwent multiple procedures during the operation were given an ACHS score with the highest mortality score. Mortality was determined as the hospital mortality.

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Table 1
Distribution of diagnoses, mortality and number of operation

			Mortality		Number of operation									
					1		2		3		4		5	
	n	%	n	%	n	%	n	%	n	%	n	%	n	%
Aortic valve repair	1	0.5	0	0	1	100	0	0	0	0	0	0	0	0
Aortic aneurysm	2	1.0	0	0	1	50	0	0	1	50	0	0	0	0
Aortic stenosis, subvalvular	1	0.5	0	0	1	100	0	0	0	0	0	0	0	0
Atrial septal defect	28	13.7	0	0	28	100	0	0	0	0	0	0	0	0
Aortic valve replacement	2	0.1	0	0	2	100	0	0	0	0	0	0	0	0
Coronary artery bypass graft	2	1.0	0	0	1	50	1	50	0	0	0	0	0	0
Double chamber right ventricle	9	4.4	0	0	7	77.8	2	22.2	0	0	0	0	0	0
Ebstein anomaly	6	2.9	1	16.7	6	100	0	0	0	0	0	0	0	0
Fontan operation	3	1.5	1	33.4	0	0	2	66.7	0	0	0	0	1	33.4
Fontan revision	1	0.5	1	100	0	0	0	0	0	0	0	0	1	100
Aortic coarctation	2	1.0	0	0	2	100	0	0	0	0	0	0	0	0
Conno procedure	1	0.5	1	100	0	0	0	0	0	0	1	100	0	0
Mitral valve repair	9	4.4	2	22.3	2	22.3	4	44.4	3	33.3	0	0	0	0
Mitral valve replacement	3	1.5	0	0	1	33.3	2	66.7	0	0	0	0	0	0
Partial atrioventricular septal defect	19	9.3	0	0	19	100	0	0	0	0	0	0	0	0
Pulmonary artery reconstruction	7	3.4	1	14.3	5	71.4	2	28.6	0	0	0	0	0	0
Pace implantation	2	1.0	0	0	1	50	1	50	0	0	0	0	0	0
Partial abnormal pulmonary venous return	26	12.7	0	0	26	100	0	0	0	0	0	0	0	0
Patent ductus arteriosus	3	1.5	0	0	3	100	0	0	0	0	0	0	0	0
Pulmonary valvuloplasty	3	1.5	0	0	3	100	0	0	0	0	0	0	0	0
Pulmonary valve replacement	15	7.3	0	0	1	6.7	11	73.3	3	20.0	0	0	0	0
Right ventricle-pulmonary artery conduit	3	1.5	1	33.4	1	33.4	1	33.3	1	33.3	0	0	0	0
Right ventricular outflow tract reconstruction	10	4.9	0	0	3	30.0	6	60.0	0	0	1	10.0	0	0
Aortopulmonary shunt	3	1.5	1	33.4	1	13.3	2	66.7	0	0	0	0	0	0
Scimitar syndrome	3	1.5	0	0	3	100	0	0	0	0	0	0	0	0
Tricuspid valve repair	7	3.4	0	0	4	57.1	3	42.9	0	0	0	0	0	0
Tetralogy of Fallot	5	2.4	0	0	5	100	0	0	0	0	0	0	0	0
Ventricular septal defect	29	14.1	0	0	21	72.4	8	27.6	0	0	0	0	0	0

Statistical analysis

The NCSS (Number Cruncher Statistical System) version 2007 (NCSS LLC., Kaysville, UT, USA) software was used for statistical analyses. Descriptive statistical methods (mean, standard deviation, median, frequency, ratio, minimum, maximum) were used to evaluate the data of the study. The compliance of the quantitative data with the normal distribution was tested via the Kolmogorov-Smirnov test, Shapiro-Wilk test, and graphical assessments. The Mann-Whitney U test was used to compare two groups that did not indicate normal data distribution. In the comparison of qualitative data, the Fisher-Freeman-Halton exact test and Fisher exact test were used. Diagnostic screening tests (sensitivity, specificity, positive predictive value, and negative predictive value) and ROC curve analysis were used to determine the cut-off value of the parameters. A *p* value of <0.05 was considered statistically significant.

RESULTS

The distribution of the patients according to diagnoses, numbers of reoperations, and mortality is presented in Table 1. The most frequent diagnoses were ventricular septal defect with 14.1% (n=29), atrial septal defect with 13.7% (n=28), partial abnormal pulmonary venous return with 12.7% (n=26), partial atrioventricular septal defect with 9.3% (n=19), and pulmonary valve replacement with 7.3% (n=15).

The ABC, STAT, and ACHS score distributions according to mortality are provided in Table 2. Mortality was observed in 4.4% (n=9) of the patients included in the study. In terms of mortality, statistically significant differences were identified for ABC, STAT (p=0.001; p<0.01), and ACHS scores (p=0.037; p>0.05); the scores of patients with mortality were higher. We used the binomial exact test (DeLong test) to compare the areas under the ROC curves. Accordingly, the ABC and STAT mortality scores were not significantly different in predicting mortality (p>0.05), whereas the ABC and ACHS scores were significantly associated (p<0.05). There was also a borderline significance between the STAT mortality score and the ACHS score (p≤0.05; Figure 1). Based on this significance, cut-off points were calculated for the scoring systems. The cut-off values and ROC curve results are given in

Table 2
Evaluation of aristotle basic, STAT and ACHS scores according to mortality

	Mortality										
	(-) (n=196)					(+) (n=9)					
	n	%	Mean±SD	Median	Q1-Q3	n	%	Mean±SD	Median	Q1-Q3	*p*
Aristotle basic complexity score			6.64±2.12	7.0	5-8			10.70±2.36	10.0	9.5-12	0.001*†
Aristotle basic complexity category											0.001*‡
Level 1	59	100				0	0				
Level 2	74	98.7				1	1.3				
Level 3	45	95.7				2	4.3				
Level 4	18	75.0				6	25.0				
STAT mortality score			0.51±0.38	0.5	0.2-0.7			1.58±0.84	1.4	0.8-2.5	0.001*†
STAT mortality category											0.001*‡
Level 1	87	100				0	0				
Level 2	78	98.7				1	1.3				
Level 3	18	85.7				3	14.3				
Level 4	13	72.2				5	27.8				
ACHS score			0.39±0.27	0.4	0.2-0.5			0.92±0.91	0.4	0.4-0.9	0.034**†

STAT: Society of Thoracic Surgery European Association for Cardiothoracic Surgery; ACHS: Adult Congenital Heart Surgery; SD: Standard deviation; Q: Quartile; † Mann Whitney U Test; ‡ Fisher Freman Halton test; * p<0.01; ** p<0.05.

Table 3
Diagnostic screening tests and ROC curve results for ABC, STAT and ACHS score by mortality

	Diagnostic scan						ROC curve	
	Cut off	A	B	PPV	NPV	AUC	95% CI	*p*
ABC score	≥9	88.89	84.69	21.05	99.40	0.899	0.784-1.000	0.001**
STAT score	≥0.8	88.89	84.18	20.51	99.41	0.898	0.790-1.000	0.001**
ACHS score	≥0.7	44.44	94.39	26.67	97.37	0.706	0.538-0.874	0.037*

ROC: Receiver operating characteristic; ABC: Aristotle Basic Complexity score; STAT: Society of Thoracic Surgery European Association for Cardiothoracic Surgery; ACHS: Adult Congenital Heart Surgery; A: Sensitivite; B: Spesifisite; PPV: Positive predictive value; NPV: Negative predictive value; AUC: Area under the curve; CI: Confidence interval; * p<0.05; ** p<0.01.

Table 3 (Figure 2). The cut-off values were ≥9, ≥0.8, and ≥0.7 for the ABC, STAT, and ACHS scores, respectively, and a statistically significant difference was observed between these values (p=0.001; p <0.01). The numbers of reoperations and postoperative complications, the mortality scores of the patients who underwent reoperation, and the relevant ROC curve results are presented in Tables 4, 5, and 6, respectively (Figure 2).

DISCUSSION

Adults with congenital heart disease make up a rapidly growing segment of the cardiovascular patient population.[2] In their study, Gilboa et al.[3] estimated that the number of patients reached 1.4 million. The incidence of congenital heart disease in Türkiye is about 1%, and there are approximately 12,000 new patients each year.[4] Today, more than 85% of children

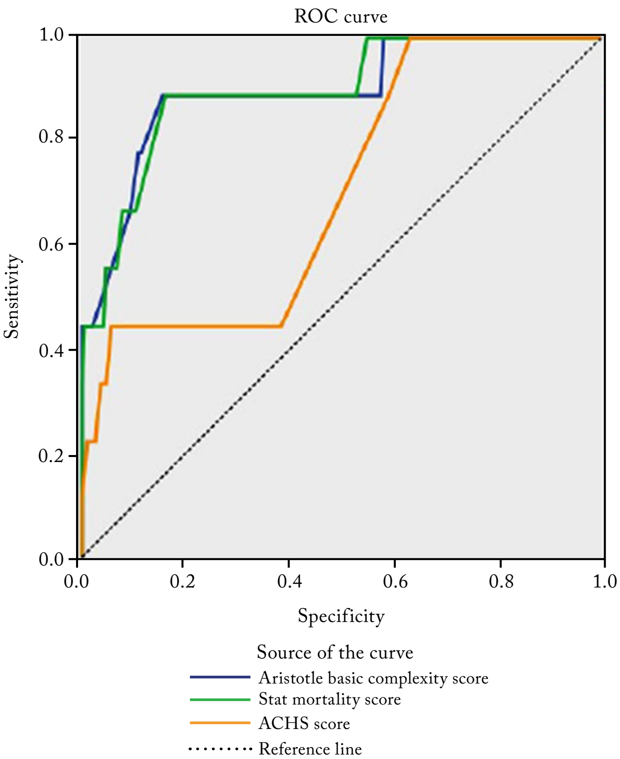

Figure 1. ROC curves for ABC, STAT and ACHS scores by mortality.

ROC: Receiver operating characteristic; ABC: Aristotle Basic Complexity score; STAT: Society of Thoracic Surgery European Association for Cardiothoracic Surgery; ACHS: Adult Congenital Heart Surgery.

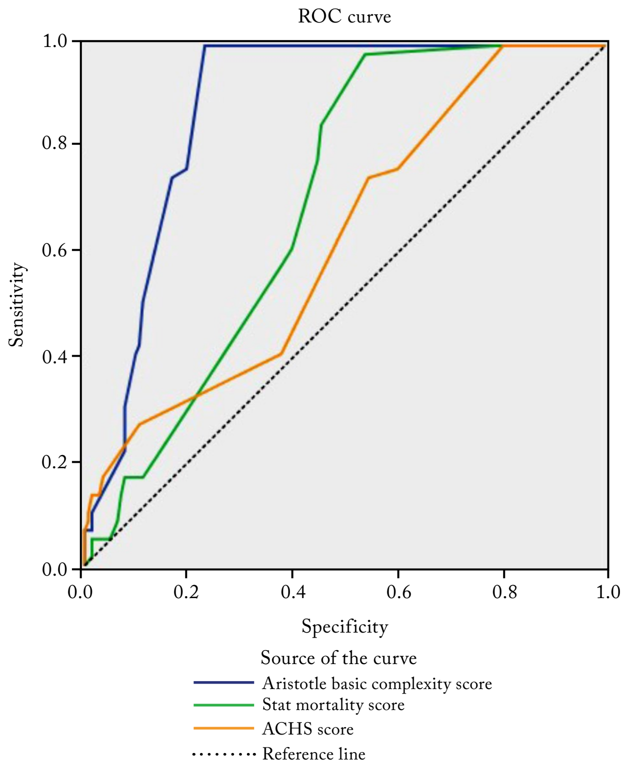

Figure 2. ROC curves for ABC, STAT and ACHS scores according to mortality in reoperated patients.

ROC: Receiver operating characteristic; ABC: Aristotle Basic Complexity score; STAT: Society of Thoracic Surgery European Association for Cardiothoracic Surgery; ACHS: Adult Congenital Heart Surgery.

Table 4
Distribution of reoperation and postoperative complication

	n	%	*p*
Reoperation			
(-)	146	71.2	
(+)	59	28.8	
2nd operation	47	22.9	
3rd operation	7	3.4	
4th operation	3	1.5	
5th operation	2	1.0	
Postoperative complication			0.02*
(-)	177	86.3	
(+)	28	13.7	
Wound infection	2	7.1	
Arrhythmia	5	17.9	
Neurological complication	3	10.7	
Delayed sternal closure	2	7.1	
Low cardiac output	4	14.3	
Postoperative bleeding-induced exploration	7	25.0	
ECMO	3	10.7	
Postoperative early reoperation	7	25.0	
Prolonged ventilation	5	17.9	
Pericardial effusion	3	10.7	
Mortality			
(-)	196	95.6	
(+)	9	4.4	

ECMO: Extracorporeal membrane oxygenation; Fisher Freeman Halton exact test; * $p<0.01$.

with congenital heart disease are expected to reach adulthood.[5,6]

Risk stratification regarding the procedures is important in determining surgical mortality. In 2004, Lacour-Gayet et al.[7] developed the ABC score to assess surgeon performance in congenital heart surgery. In 2006, Kang et al.[8] demonstrated that this score is somewhat useful in determining mortality but cannot be used as a statistically significant tool. In 2007, O'Brien et al.[9] argued that the ABC score is useful for differentiating low-and high-risk patients. A 2011 study by Photiadis et al.[10] supported this opinion but indicated that the ABC score is based on the complexity of surgical procedures and does not take into account patient-related factors. In their 2012 study, Hörer et al.[11] demonstrated that the ABC score is not suitable for predicting mortality in combined surgical procedures.

In 2014, Kogon et al.[12] published a study comparing the STAT and ABC scores. In this retrospective study, they argued that the scoring systems used in pediatric patients were also valuable in the adult group. They suggested that ABC and STAT scores had similar results in determining mortality; however, ABC score was more effective in determining major complications and length of hospitalization.

In 2015, Cavalcanti et al.[13] argued that the ABC and STAT scores were not significantly different in predicting mortality. A 2016 study by Hörer et al.[14] suggested that the ABC score had a low predictive value but outperformed the STAT score. In 2019, Bobillo-Perez et al.[15] indicated that the predictive performance of the STAT score was better than that of the ABC score.[16]

The controversial state of scoring systems in the adult group caused the search for a new scoring system. In their study published in 2015, Fuller et al.[1] suggested that the ACHS mortality score was effective. In this study, they divided the patients into 152 groups and determined an ACHS mortality score

Table 5
Evaluation of ABC, STAT and ACHS scores according to mortality in reoperated patients

	Mortality										
	(-) (n=146)					(+) (n=59)					
	n	%	Mean±SD	Median	Q1-Q3	n	%	Mean±SD	Median	Q1-Q3	*p*
Aristotle basic complexity score			6.64±2.12	7.0	5-8			10.70±2.36	10.0	9.5-12	0.001*†
Aristotle Basic Complexity category											0.001*‡
Level 1	59	100				0	0				
Level 2	60	80.0				15	20.0				
Level 3	16	34.0				31	66.0				
Level 4	11	45.8				13	54.2				
STAT mortality score			0.51±0.38	0.5	0.2-0.7			1.58±0.84	1.4	0.8-2.5	0.001*†
STAT mortality category											0.001*‡
Level 1	78	89.7				9	10.3				
Level 2	44	55.7				35	44.3				
Level 3	14	66.7				7	33.3				
Level 4	10	55.6				8	44.4				
ACHS score			0.39±0.27	0.4	0.2-0.5			0.92±0.91	0.4	0.4-0.9	0.034**†

ABC: Aristotle Basic Complexity score; STAT: Society of Thoracic Surgery European Association for Cardiothoracic Surgery; ACHS: Adult Congenital Heart Surgery; SD: Standard deviation; Q: Quartile; † Mann Whitney U test; ‡ Pearson Chi square test; * $p<0.01$; ** $p<0.05$.

ranging from 0.1 to 3.0 for each. In 2019, Abouelella et al.[17] stated that the ACHS score is currently the best predictor of mortality in GUCH patients and that the mortality rate was 4%.

In our study, we investigated the effectiveness of GUCH using this scoring system. In the present study, hospital mortality was 4.4% (n=9). In terms of mortality, statistically significant differences were identified for ABC, STAT ($p=0.001$; $p<0.01$), and ACHS scores ($p=0.037$; $p>0.05$); the scores of patients with mortality were higher (Table 3). In our study, we calculated the cut-off values and performed ROC curve analysis for the prediction of mortality. The ROC curve results for ABC, STAT, and ACHS scores for predicting mortality are given in Table 3. Based on this significance, cut-off points were calculated for the scoring systems. The incidence of mortality was 13.455 times higher in patients with a cut-off value of ≥0.7 in their ACHS mortality scores.

Abouelella et al.[17] reported a postoperative complication rate of 18%, similar to the rate of 28% stated by Mascio et al.[18] The reported incidence of neurological complications is 7%.[19] In our study, postoperative complications were detected in 13.7% of the cases (n=28), and it is consistent with the literature (Table 4). In our study, extracorporeal membrane oxygenation was required in 10.7% (n=3) of patients (aortopulmonary shunt, n=1; pulmonary artery reconstruction, n=1; mitral valve repair, n=1) due to low cardiac output. All of these patients died while on support. Neurological complications were observed in 10.7% (n=3) of our patients, including one patient who underwent right ventricle to pulmonary artery conduit replacement and later died due to intracranial bleeding, as well as two patients who underwent pulmonary valve replacement and mitral valve replacement. These patients developed postoperative convulsions, but no pathologies were detected in their examinations; therefore, the condition was attributed to temporary ischemia and completely resolved with medical treatment.

A statistically significant relationship was found between postoperative complication rates according to the occurrence of reoperation ($p=0.002$; $p<0.01$); the rate of postoperative complication incidents in those reoperated was higher than in those who were not reoperated. The ABC, STAT, and ACHS results of reoperated patients are demonstrated in Table 5.

Table 6
ROC curve results for ABC, STAT and ACHS scores in reoperated patients

	ROC curve		
	AUC	95% CI	*p*
Aristotle basic complexity score	0.881	0.835-0.927	0.001*
STAT mortality score	0.698	0.628-0.768	0.001*
ACHS score	0.626	0.544-0.707	0.005*

AUC: Area under the curve; ROC: Receiver operating characteristic; CI: Confidence interval; * p<0.01.

The ABC, STAT, and ACHS scores of the patients who underwent reoperation were significantly higher than the scores of those who did not. The areas under the ROC curves were 0.81 (p=0.001, p<0.01), 0.69 (p=0.001, p<0.01), and 0.62 (p=0.034, p<0.05), respectively.

When the areas undser the ROC curve were compared, the ABC score was found to be more effective in predicting mortality than the ACHS score (p=0.001; p<0.01). The STAT mortality score and ACHS score were not significantly different (p=0.626; p>0.05; Table 6).

The small sample size of the study is its main limitation. Some complex surgeries, such as Fontan operation, are rare in adulthood, and therefore the number of patients to be compared is small. Additionally, some data losses are not excluded due to the retrospective design.

In conclusion, for primary operations, all scoring systems could significantly predict mortality; however, the ABC and STAT scores had better predictive value compared to the ACHS score. The predictive value of STAT and ACHS scores was similar in reoperations, whereas the ABC score had a higher predictive value. The ACHS mortality score has good predictive power in adult congenital heart patients. Preoperative risk prediction could be used safely to analyze surgical results.

Ethics Committee Approval: The study protocol was approved by the Koşuyolu High Specialization Education and Research Hospital Ethics Committee (Date: 08.12.2020, no: 2020/13/390). The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patient Consent for Publication: A written informed consent was obtained from each patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept, design:B.Z.T.R.; Control/supervision: H.C., E.T.; Data collection and/or processing, analysis and/or interpretation: B.Z.T.R.; Literature review: B.Z.T.R., E.T.; Writting the article: B.Z.T.R.; Critical review: B.Z.T.R., H.C.; References and funding, materials: N.C.

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