## Comparison of risk stratification scoring system as a predictor of mortality and morbidity in congenital heart disease patients requiring surgery

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#### ABSTRACT

Backgrounds	:	Risk stratification systems have been important in reducing morbidity and mortality among congenital heart disease (CHD) patients requiring cardiac surgery. Multiple risk stratification scoring systems have been developed, including Aristotle Basic Complexity Score (ABC), Aristotle Comprehensive Complexity Score (ACC), Society of Thoracic Surgeons and European Association for Cardiothoracic Surgery (STS-EACTS), and Risk Adjustment in Congenital Heart Surgery (RACHS-1). This study aims to access the superior risk stratification scoring system model in predicting mortality and morbidity.
Methods	:	The authors used Embase, PubMed, Scopus, and ProQuest as the primary databases for searching and included studies from hand searching. The area under the receiver operating characteristic curve was compared.
Results	:	A total of 11 articles were included in this review. The AUC of ABC for predicting mortality ranges from 0.59 to 0.71, and morbidity ranges from 0.673 to 0.743. The AUC of ACC score for predicting mortality ranges from 0.704 to 0.87, and a study revealed the AUC of morbidity is 0.730. The AUC of RACHS-1 for predicting mortality ranges from 0.68 to 0.782. The AUC of STS-EACTS for predicting mortality ranges from 0.739 to 0.8 and 0.732 for predicting morbidity.
Conclusion	:	ABC, ACC, RACHS-1, and STS-EACTS have acceptable to excellent discriminatory ability in predicting mortality and morbidity among CHD patients requiring cardiac surgery.
Keywords	:	Aristotle Basic Complexity Score, congenital heart surgery, risk adjustment in congenital heart surgery-1, Society of Thoracic Surgeons and European Association for Cardiothoracic Surgery

## **INTRODUCTION**

Congenital heart disease (CHD) is the most frequently occurring congenital defect, affecting millions of neonates yearly. Although there is a significant increase in the prevalence of CHD, there is also a notable

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decrease in the mortality rate in CHD patients who underwent surgery. Some of the most important causes of this improvement are early detection, adequate risk

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assessment, and improvement in surgical techniques. Risk stratification tools have played an important role in reducing the mortality and morbidity of CHD patients requiring surgery.<sup>[1-3]</sup> There are currently multiple risk stratification tools available, including Aristotle Basic Complexity Score (ABC), Aristotle Comprehensive Complexity (ACC), Risk Adjustment in Congenital Heart Surgery (RACHS-1), Society of Thoracic Surgeons and European Association for Cardiothoracic Surgery (STS-EACTS), and many others. ABC score is a consensus-based risk stratification tool; it calculates and ranks the risk of the surgery based on mortality potential, morbidity potential, and anticipated technical difficulty.<sup>[4]</sup> Meanwhile, ACC further adjusted the complexity by incorporating the characteristics of the patients, including anatomical factors, related procedures, comorbidity, and other factors.<sup>[5]</sup> RACHS-1 was made according to the categorization of multiple procedures, including palliative and corrective surgery, with a similar mortality rate. RACHS1 stratifies CHD surgery into six categories.<sup>[6]</sup> One of the newer risk stratification systems is the STS-EACTS score. Introduced in 2008, STS-EACTS was made with objective data and minimal use of subjective probability.<sup>[7]</sup> The availability of multiple risk stratification systems provides clinicians with options to choose the most suitable system for their hospital usage. However, it also raises a question about which risk stratification system is superior to the other and which one is most applicable to a different clinical setting. This systematic review aims to find the superior risk stratification system, especially in predicting mortality and morbidity in CHD patients.

## **METHODS**

The protocol of this article has been registered at Prospero (Registration ID: CRD42023415919). The protocol and data extraction were conducted in accordance with the 2009 PRISMA guideline (completed checklist available upon request). Articles will be obtained from PubMed, Scopus, Embase, ProQuest, and through hand search.

### Study selection

Articles were eligible if they compared at least two scoring systems: ABC, ACC, RACHS-1, and STS-EACTS. The outcome of these articles was mortality, defined by deaths occurring during the in-hospital stay or 30 days postsurgery, and morbidity, defined by length of stay. The article should include CHD patients requiring surgery. Duplicates, articles with no online full text available, case reports, case series, editorial letters, expert opinions, and conference abstracts are excluded from this study.

#### Data extraction and analysis

The extracted results from various searches were independently analyzed by the authors. In the event of any disagreements, they were resolved through thoughtful discussions and reaching a consensus. The data compiled for this study include the area under the receiver operating characteristic curve (AUROC), which represents the scoring system's discriminatory power (i.e., capacity to identify patients at higher risk of meeting the outcome), and its calibration, which is represented by the *P* value of the Hosmer-Lemeshow goodness-of-fit test. AUROC values >0.8 were regarded as having a high discriminatory power.

#### **Risk of bias**

The assessment of risk of bias (ROB) was conducted for each study using the Prediction Model Risk of Bias Assessment Tool (PROBAST). The tool has four distinct areas, namely, participants, predictors, results, and analysis. Within these categories, a comprehensive set of 20 signaling questions is provided to aid in the evaluation of the ROB. In addition to evaluating the ROB, the applicability of the model was assessed to determine whether the participants, predictors, and outcomes were relevant to the research question.

## RESULTS

An initial search retrieved 1245 articles. Out of these, 231 articles were detected to be duplicates and were removed. Further, 984 articles were then reviewed and excluded. Thirty articles were selected for further review, and 15 articles were excluded because they only analyzed one risk stratification scoring system and/or did not provide adequate information. Four articles were excluded because of the publication type. Therefore, a total of 11 articles were included in this article.

#### Outline of the included studies

A total of 20,812 patients were included in the selected studies is shown in Table 1. Ranging from the smallest sample size of 360 patients to the largest of 11,438 patients. One study compares ABC, ACC, RACHS-1, and STS-EACTS; three studies compare ABC, RACHS-1, and STS-EACTS; three studies compare ABC, ACC, and RACHS-1; and four studies compare ABC and RACHS-1.

#### Aristotle basic complexity

The AUC for predicting mortality ranges from 0.59 to 0.71, suggesting low-to-acceptable discriminatory ability. However, these numbers are still lower when compared to the AUC for predicting mortality from other scoring systems. A study by Sabuncu *et al.* compares ABC, ACC, and RACHS-1 by the Mann–Whitney *U* test and found that

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ABC has the lowest ability to predict mortality compared to the other two.<sup>[8]</sup> Among 11 studies, three observed the ability of ABC to predict morbidity. The AUC of ABC for predicting morbidity ranges from 0.673 to 0.743. These AUC values suggest acceptable discriminatory ability but are still lower than the other scoring systems observed in the included studies.<sup>[2-4]</sup>

#### Aristotle comprehensive complexity

The AUC of ACC score for predicting mortality ranges from 0.704 to 0.87, suggesting acceptable-to-excellent discriminatory ability.<sup>[2,5,6]</sup> A study by Sabuncu *et al.* comparing ABC, ACC, and RACHS-1 by the Mann-Whitney U test showed that ACC could predict mortality most sharply compared to the other two, especially for the first 10 days post-surgery.<sup>[8]</sup> From the included studies, only one study observed the ACC score's ability to predict morbidity. A study by Yildiz *et al.* found that the AUC of ACC score for predicting morbidity is 0.730, which suggests acceptable discriminatory ability.<sup>[9]</sup>

#### Risk adjustment in congenital heart surgery 1

The AUC of RACHS-1 for predicting mortality ranges from 0.68 to 0.782, suggesting acceptable discriminatory ability.<sup>[1-7,9,10]</sup> When compared to the ABC score, the results are varied. A study by Al-Radi et al. comparing ABC and RACHS-1 showed that the difference between the c-index of ABC and RACHS-1 models was significant (P = 0.018, c-index 0.698 vs. 0.733, respectively) and that the predictive value of RACHS-1 for morbidity was higher than that of ABC (P < 0.0001).<sup>[11]</sup> This finding is supported by the study comparing ABC and RACHS-1 by Lelong et al. Their study found that the ABC score has a lower discriminatory ability than RACHS-1 (P = 0.003).<sup>[4]</sup> However, a study by Alam et al. found that the RACHS-1 model has the worst predictive value for both prolonged PLOS (0.701) and hospital mortality (0.766) when compared to ABC and STS-EACTS.<sup>[12]</sup>

# Society of Thoracic Surgeons and European Association for Cardiothoracic Surgery

The AUC of STS-EACTS for predicting mortality ranges from 0.739 to 0.8, suggesting acceptable-to-excellent discriminatory ability. As shown in the four included studies, the STS-EACTS system shows the highest AUC compared to other scoring systems. However, only one study found statistical significance.<sup>[2-4,7]</sup> The study comparing ABC, RACHS-1, and STS-EACTS by Alam *et al.* found a statistically significant difference between the AUC of ROC curves of STS-EACTS and RACHS-1 model for both morbidity (P = 0.046) and hospital mortality (P = 0.015). The AUC for predicting morbidity ranges from 0.732 to 0.759, suggesting acceptable discriminatory ability.<sup>[12]</sup> A study comparing ABC, RACHS-1, and STS-EACTS by Bobillo-Perez *et al.* also found a statistically significant difference between

study	Population			MORTAILLY			MOR	Morbiaity	
		ABC	ACC	RACHS-1	STS-EACTS	ABC	ACC	RACHS-1	STS-EACTS
Lelong <sup>[4]</sup>	443 patients	AUC=0.59 95%CI=0 49-0 69	I	AUC=0.68.95% 95%CI=0.58-0.79					
Cavalcanti et al.⊡	360 patients	AUC=0.766 95%CI=0.718-0.808	·	AUC=0.738 95%Cl=0.690-0.783	AUC=0.738 AUC=0.739 5%Cl=0.690-0.783 95%Cl=0.691-0.784				
Yildiz <i>et al.</i> <sup>[9]</sup>	1,950 patients	AUC=0.712 95%CI=0.668-0.755	AUC=0.795 95%CI=0.753-0.837	AUC=0.729 95%CI=0.689-0.770	AUC=0.712 AUC=0.795 AUC=0.729 AUC=0.803 AUC=0.685 AUC=0.730 AUC=0.731 AUC=0.732 95%Cl=0.689-0.711 95%Cl=0.755 95%Cl=0.756 95%Cl=0.707-0.756	AUC=0.685 95%CI=0.659-0.711	AUC=0.730 95%Cl=0.705-0.755	AUC=0.731 95%CI=0.706-0.755	AUC=0.732 95%CI=0.707-0.756
Bobillo-perez <i>et al.</i> <sup>[10]</sup>	1,037 patients	AUC=0.658	T	AUC=0.760	AUC=0.763	AUC=0.673	F	AUC=0.714	AUC=0.733
Alam <i>et al.</i> <sup>[12]</sup>	920 patients	AUC=0.817 95%CI=0.717-0.916		AUC=0.766 95%CI=0.657-0.874	AUC=0.766 AUC=0.870 AUC=0.743 95%Cl=0.657-0.874 95%Cl=0.824-0.915 95%Cl=0.717-0.916	AUC=0.743 95%CI=0.717-0.916		AUC=0.766 95%CI=0.657-0.874	AUC=0.766 AUC=0.759 95%CI=0.657-0.874 95%CI=0.720-0.797
DeCampli et al. <sup>[13]</sup>	1,103 patients	AUC=0.63 05%CI=0.55.0.72	AUC=0.81 95%CI=0.55. 0.72	AUC=0.73 95%CI=0.66-0.80					
Sabuncu et al. <sup>[8]</sup>	413 patients	Z=-6.557	Z=-9.102	Z=-7.782					
t al.[ <sup>14]</sup>	1,150 patients	AUC=0.677 95%CI=0.61-0.73	AUC=0.704 95%CI=0.64-0.76	AUC=0.607 95%CI=0.55-0.66	ı				
Carmona <sup>[15]</sup> Al-Badi <i>et al</i> [11]	Carmona <sup>[15]</sup> 614 patients Al-Radi <i>et al</i> <sup>[11]</sup> 11 438 natients	AUC=0.650 C-index=0.018		AUC=0.699 C-index=0.733					
Bojan <i>et al.</i> <sup>[16]</sup>	1,384 patients		c-indexes 0.87 (0.84. 0.91)	C-index=0.75 (0.65. 0.82)					

STS-EACTS and ABC, but no statistically significant difference between STS-EACTS and RACHS-1.<sup>[10]</sup>

#### **Risk of bias**

Based on a review using PROBAST on research participants, research objectives, outcomes, and analysis of the literature used, ROB is stated to be a low ROB. All predictive models are made with external validation and have large enough data. There was low concern regarding the applicability of models for participants, predictors, and outcomes.

## **DISCUSSION**

This systematic review identified and evaluated four risk stratification scoring systems as a predictor of mortality and morbidity in CHD patients requiring surgery.

The ABC score is a comprehensive metric utilized to assess the case complexity of congenital heart surgery procedures. This scoring system incorporates three fundamental components, namely, the potential for mortality, the potential for morbidity, and the technical intricacy associated with the procedure. By considering these crucial factors, the ABC score provides a standardized approach to evaluating the complexity of congenital heart surgeries.<sup>[13]</sup> The comprehensive ABC score spans from 1.5 to 15 points, with higher ratings denoting increased overall complexity. The processes were categorized based on the score into four levels: Level 1 (1.5-5.9), Level 2 (6.0-7.9), Level 3 (8.0-9.9), and Level 4 (10.0–15.0). The scoring system was established by a consortium of 50 surgeons hailing from 23 different nations in the year 1999.<sup>[12]</sup>

The ACC scoring system is a system that is based on the ABC scoring system. The scoring system incorporates patient-related elements inside its framework. The factors under consideration can be categorized into two distinct groups: those that are directly associated with the procedure itself and those that are unrelated to the procedure. The ACC scoring system is comprised the summation of the ABC scoring system and the scores of the patient-specific factors. In the ACC scoring system, the maximum possible score is increased from 15 to 25, and the complexity level is raised from 4 to 6 (Level 5 = 15.1-20.0 and level 6 = 20.1-25.0). In the ACC scoring system, factors that are unrelated to the surgical procedure to be performed and those that the patient had previously and that can influence the patient's mortality and morbidity risk independently of the surgical procedure are included.<sup>[8]</sup> In 1997, the Children's Hospital Boston team assembled a commission of 11 nationally representative pediatric cardiologists and cardiac surgeons to develop the RACHS-1 method.<sup>[7,8]</sup> The RACHS-1 system utilizes a scale that spans from one to six. The RACHS-1 scoring system enables the incorporation

of variables including age, preterm, and noncardiac congenital anatomical anomalies, hence facilitating adjustments for these parameters.<sup>[9]</sup> The STS and the EACTS mortality score, which was developed in 2008, represent the most recent systems in this field. The study utilized a dataset consisting of real-world data from a total of 77,294 patients (33,360 patients from the EACTS and 43,934 patients from the STS) who had 148 different types of procedures. The time frame for data collection spanned from 2002 to 2007. The primary objective of the study was to assess the mortality risk associated with each of these treatment types. Mortality rates were computed for each procedure utilizing Bayesian statistics that are tailored to accommodate data with tiny denominators. The scale exhibits a range spanning from 0.1 to 5.0, with an associated categorization of mortality levels ranging from 1 to 5. These levels are allocated as follows: Level 1 corresponds to a range of 0.1–0.3, level 2 corresponds to a range of 0.4–0.7, level 3 corresponds to a range of 0.8-1.2, level 4 corresponds to a range of 1.3-2.6, and level 5 corresponds to a range of 2.9-5.0.<sup>[7,8]</sup>

Our search only found one article that compared all four systems. This study by Yildiz et al. suggests that STS-EACTS provided a superior prediction of mortality and morbidity. However, they found no statistically significant differences between the four systems' areas under the curve.<sup>[9]</sup> This finding was also supported by studies by Cavalcanti et al., Bobillo-Perez et al., and Alam et al. Alam et al. compared ABC, RCHS-1, and STS-EACTS in 2018. Their study found statistical differences between the AUC of ROC curves of the STS-EACTS and RACHS-1 model for both morbidity (P = 0.046) and hospital mortality (P = 0.015).<sup>[7,10,12]</sup> Bobillo-Perez et al. found a statistically significant difference between STS-EACTS and ABC, but no statistically significant difference between STS-EACTS and RACHS-1. We found three studies that compare ABC, ACC, and RACHS-1.<sup>[10]</sup> These three studies by DeCampli et al., Sabuncu et al., and Joshi et al. similarly conclude that ACC can better predict mortality in patients with CHD requiring surgery. There are three studies comparing ABC and RACHS-1.<sup>[13,14]</sup> Studies by Lelong et al., Carmona et al., and Al-Radi et al. all conclude that RACH-1 has a better ability to predict mortality than ABC.[4,11,15] Al-Radi et al. also found that the difference between the c-index of ABC and RACHS-1 models was significant (P = 0.018).<sup>[11]</sup>

## **CONCLUSIONS**

This systematic review re-established the usability of ABC, ACC, RACHS-1, and STS-EACTS in predicting mortality and morbidity. There are different findings across the studies on deciding the best risk stratification scoring system. Choosing the superior model will require adjustment according to the centers and patients that will be involved. Further research with large prospective studies is required to validate these findings.

#### Data availability

All data generated or analyzed during this study are included in this published article and its supplementary information files.

#### Authors' contributions

Study conception and design: Dicky Fakhri; Data collection: Ni Made Ayu Sintya Damayanti; Analysis and interpretation of results: Dicky Fakhri, Muhammad Nurhanif; Draft manuscript preparation: Ni Made Ayu Sintya Damayanti, Muhammad Nurhanif. All authors reviewed the results and approved the final version of the manuscript.

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Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

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