Surgical Management of Aortic Coarctation from Infant to Adult

Infantdan Yetişkine Aort Koarktasyonun Cerrahi Tedavisi

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ABSTRACT

Objective: In the present study, we aimed to retrospectively investigate the early and late results of different surgical treatment techniques applied in different age groups with coarctation of the aorta (CoA).

Materials and Methods: Between January 2007 and February 2017, 26 patients (12 males, 14 females; mean age: 12.2±12.4 years; range: 29 days–34 years) who underwent surgery with the diagnosis of CoA were evaluated. Overall, 11 of these patients (42.3%) were in the infantile period, whereas 15 patients (57.7%) aged between 6 and 34 years. Resection and end-to-end anastomosis were performed in 13 patients (50%). Bypass grafting was performed in six patients (23.1%), and patch plasty was performed in seven patients (26.9%).

Results: A patient (3.8%) who was operated on during the infantile period died early, whereas another patient (3.8%) died 2 years after the surgery. Recoarctation was detected in two patients. A patient underwent balloon dilatation, whereas another patient underwent balloon dilatation and stenting. In patients who underwent resection and end-to-end anastomosis based on postoperative echocardiography results during follow-up, a lower statistically significant gradient was observed compared with the preoperative period. Despite the decrease in the left ventricular systolic diameter (LVSD) and the increase in the ejection fraction (EF) the decrease in LVSD and increase in EF were not statistically significant. In patients who underwent patch plasty or graft interposition, the low values of the gradient and left ventricular diastolic diameter in the postoperative follow-up were statistically significant. However, the decrease in LVSD and increase in EF were not statistically significant.

Conclusions: Our clinical experience suggests that repairing with resection and end-to-end anastomosis is a more appropriate treatment option during the infancy, whereas patch plasty or bypass grafting may be preferred in advanced ages.

Keywords: Coarctation of the aorta, surgery, adult

ÖZ

Amaç: Bu çalışmada aort koarktasyonlu farklı yaş gruplarında uygulanan farklı cerrahi tedavi tekniklerinin erken ve geç dönem sonuçları geriye dönük incelenmesi amaçlandı.

Gereç ve Yöntem: Ocak 2007 ile Şubat 2017 yılları arasında aort koarktasyonu tanısı ile ameliyat edilen 26 hasta (12 erkek, 14 kadın, ort yaş: 12,2±12,4; range 29 gün-34 yıl) değerlendirildi. Hastaların 11 (%42,3) tanesi infantil dönemdeydi, fakat 15(%57,7) hasta 6-34 yaş arasında idi. Rezeksiyon ve uç uca anastomoz 13 (%50) hastada uygulandı. Greft baypas 6 (%23,1) hastada uygulandı ve 7 (%26,9) hastada patch plasty yapıldı.

Bulgular: İnfantil dönemde ameliyat edilen 1 (%3,8) hasta erken dönemde, bir diğer hasta (%3,8) operasyondan 2 yıl sonra kaybedildi. Rekoarktasyon 2 hastada tespit edildi. Bir hastaya balon dilatasyon uygulandı, diğer hastaya balaon dilatasyon ve stent uygulandı. Hastaların postoperatif ekokardiyografi takiplerinde resection and end-to-end anastomosis yapılan hastalarda preoperatif döneme göre gradiyent düşüklüğü istatistiksel olarak anlamlı idi. Sol ventrikül sistolik çapta azalma (LVSÇ) ve Ejeksiyon fraksiyonunda (EF) artma tespit edilmiş olmasına rağmen LVSÇ düşme ve EF artış istatistiksel açıdan anlamlı değildi. Patch plasty veya graft interposition yapılan hastalarda postoperatif takiplerde gradiyent ve sol ventrikül diyastolik çap düşüklüğü istatistiksel olarak anlamlı idi. Preoperatif döneme göre LVSÇ düşme ve EF artış istatistiksel açıdan anlamlı değildi.

Sonuç: Bizim klinik tecrübelerimiz infant döneminde rezeksiyon ve uç-uca anastomoz ile tamir, daha ileri yaşlarda ise patch plasti veya greft bypass ile tamirin daha uygun tedavi seçeneği olabileceğini düşündürmektedir.

Anahtar Kelimeler: Aort koarktasyonu, cerrahi, yetişkin

Introduction

Coarctation of the aorta (CoA) is the discrete narrowing of the aorta and is the sixth most common lesion occurring in congenital heart diseases. Its prevalence is approximately 5%–8% among all congenital heart diseases. In untreated patients, complications such as hypertension, heart failure, intracranial bleeding, aortic rupture, infective endocarditis, and myocardial infarc-

tion may develop, and these complications can cause high mortality and morbidity [1]. In the most recent large and multicenter studies, the surgical mortality rate was reported as 2.4%, whereas the overall postoperative complication rate was 36%. Survival rates following CoA repair were reported as 93.3%, 86.4%, and 76.5% at 10, 20, and 30 years, respectively [1, 2]. The surgical management for CoA has changed considerably since the first successful surgical repair performed in the mid-1940s. Throughout the many decades since the first successful surgery, techniques included resection and end-to-end anastomosis, subclavian flap aortoplasty, patch graft aortoplasty, and bypass graft insertion across the area of coarctation when the distance to be bridged is extremely long for an end-to-end repair [2]. Currently, endovascular therapy (either stent implantation or balloon angioplasty) is accepted as an alternative to surgery [1].

In the present study, we investigated the early and late results of different surgical treatment techniques applied in different age groups with CoA.

Materials and Methods

An approval from the local ethics committee was obtained for this study. Written informed consent was obtained from each patient. The study was conducted in accordance with the principles of the Declaration of Helsinki. Demographic information, postoperative data, and operative details were retrospectively collected from medical records and the hospital electronic record system.

Between January 2007 and February 2017, 26 patients (12 males, 14 females; mean age: 12.2±12.4 years; range: 29 days–34 years) who underwent surgery with the diagnosis of CoA were evaluated. Overall, 11 of these patients were in the infantile period and 15 patients aged between 6 to 34 years. The indications for the surgery were according to the American Heart Association guidelines. Patient characteristics, comorbidities, symptoms and signs, associated cardiac diseases, types of operative repair, and postoperative complications were collected (Table 1).

Table 1. Preoperative details

All patients	n=26	Mean±SD	%
Age (years)		12.2±12.4	
Sex			
Male	12		46.2
Female	14		53.8
SBP (mmHg)		160±18.4	
DBP (mmHg)		95±11.6	
Gradient (mmHg)		58.2±19.4	
LVEDD (mm)		33.5±16.2	
LVESD (mm)		21.2±10.4	
LVEF (%)		65±7.4	
Recurrent coarctation after balloon			
Comorbidities			
Hypertension	8		30.8
Number of antihypertensive medications		1.2±1	
Cerebrovascular accident	2		7.7
COPD	3		11.5
Aortic or mitral valve disease	7		27
PHT	4		15.4
Symptom			
Asymptomatic	4		15.4
Claudication/limb pain	6		23.1
Chest pain	8		30.8
Headache	5		19.2
Vertigo/dizziness	3		11.5
Palpitations	8		30.8
Dyspnea	7		27
Fatigue	10		38.5
Associated anomalies			
Bicuspid aortic valve	8		30.8
Aortic dilatation	10		38.5
ASD	6		23
VSD	2		7.7
PDA	5		19.2
MVP	1		3.8
Subaortic membrane	2		7.7
Hypoplastic arch	2		7.7
Turner syndrome	1		3.8
Aberrant RSA	1		3.8

SBP: systolic blood pressure; DBP: diastolic blood pressure; LVEDD: left ventricular end diastolic diameter; LVESD: left ventricular end systolic diameter; LVEF: left ventricular ejection fraction; PHT: pulmonary hypertension; COPD: chronic obstructive pulmonary disease; ASD: atrial septal defect; VSD: ventricular septal defect; PDA: patent ductus arteriosus; MVP: mitral valve prolapse; RSA: right subclavian artery; SD: standard deviation

Renal failure was defined as the need for hemodialysis, respiratory failure was defined as the need for re-intubation or tracheostomy, and bleeding was defined as the need for re-operation. All patients were controlled by Transthoracic echocardiography (TTE), on Day 10th, and in the 1st, 3rd, and 6th month after discharge; and annually thereafter. TTE, and when needed computed tomography (CT) (Siemens, Germany) scans were obtained during follow-up. The follow-up period ranged from 1 to 10 years with a median of 5.6 years.

Statistical analysis was performed using the Statistical Package for Social Sciences (SPSS) version 13.0 software (SPSS Inc., Chicago, IL, USA). Descriptive data were expressed as mean±standard deviation or number of patients and percentage. The difference between different surgical techniques for nominal data was analyzed using the Chi-square test. Continuous variables were assessed using the student t test and Mann–Whitney U tests. A p value of <0.05 was considered statistically significant.

Operative techniques

A standard left posterolateral thoracotomy via the fourth or fifth intercostal space was per-

formed to expose the distal aortic arch. All the collaterals in the pathological segment were carefully ligated or controlled using a silicone loop. The left lung lobe was pulled to the anterior side. After the posterior pericardium was opened, the coarcted segment was reached and explored. An appropriate surgical technique was selected according to the length of the coarcted segment, its localization with ductus, age of the patient, and surgeon's preference. Patent ductus arteriosus (PDA) was closed by ligation or division method in the patients with PDA.

In patients undergoing patch aortoplasty, the aorta was suspended proximally and distally. The proximal and distal clamps were inserted and the coarctation was opened longitudinally. A posterior membrane resection was only performed when necessary. A polytetrafluoroethylene (PTFE) or Dacron® graft was used as the patch material (Figure 1). The procedure was completed using continuous propylene sutures.

Patients who were not eligible for patch aortaplasty underwent a bypass technique of the lateral isthmus with a tubular graft (Figure 2). The procedure was performed with partial cross-clamping from the proximal descending aorta to the distal descending aorta. In patients who underwent graft interposition, the coarcted segment was resected and the distal aorta tubular graft was inserted from the proximal aorta using continuous sutures.

In patients who underwent resection and end-to-end anastomosis, the coarcted segment was removed and anastomosis was performed using 5-0 and 6-0 sutures anteriorly and posteriorly with continuous sutures to prevent the narrowing of the anastomosis line and recoarctation (Figure 3).

Intercostal arteries were not routinely re-implanted into the resected segment. During the process of proximal anastomosis, minimal cooling down (32°C–34°C) was allowed for the patients with a proximal cross-clamp. Distal anastomosis was performed during re-warming of the patient.

Results

Of the 26 patients, 15 (57.7%) were diagnosed with CoA in adulthood. Systemic hypertension was the most common symptom of CoA. The mean number of antihypertensive agents used in the preoperative period, including beta blockers, calcium channel antagonists, angiotensin receptor blockers, and alpha blockers, when necessary, was 1.2±1 (range: 1–4). Femoral arterial pulses were not detected manually (palpitation by hand) in 12 of the patients (46.2%). The mean preoperative gradient was 58.2±19.4 mmHg (Table 1). Continuous murmurs were heard in 12 patients (46.2%), whereas systolic murmurs were heard in ten patients (38.5%). Congestive heart failure was observed in 13 patients (50%), whereas left ventricular hypertrophy was detected in 20 patients (76.9%). A costal notch in 16 patients was detected by telecardiography (61.5%). As additional imaging modalities, aortography, CT, and magnetic resonance angiography were performed in eight (30.8%), five (23%), and ten patients (38.4%), respectively. Presence of bicuspid aorta was the most common concomitant cardiac pathological finding. Aortic dilatation was also found in ten patients (38.4%).

Of the 11 pediatric patients, eight (72.7%) who were intervened during the infantile period underwent preoperative balloon dilatation. Recurrent CoA developed in six of these patients (75%), whereas two (25%) underwent urgent surgery because of the extravasation of guide wire and bleeding during balloon dilatation. Resection and end-to-end anastomosis were performed in 13 patients (50%): two of them were 6 and 11 years old, whereas the remaining patients were in the infantile age group. Bypass grafting was performed in six patients (23.1%), and patch plasty was performed in seven patients (26.9%). The mean hospital stay was 8.4±4.6 (range: 4–13) days. A patient (3.8%) who was operated on during the infantile period died in the early, whereas another patient (3.8%) died 2 years after the surgery. These two patients who were operated on were excluded because their postoperative data could not be obtained. Baseline characteristics of the patients are shown in Table 2.

Two of the patients with aortic dilatation underwent the modified Bentall procedure 1 and 4 years after the repair of CoA. In a patient, the modified Bentall procedure +patch plasty was performed with a simultaneous two-stage operation. In a 33-year-old man with an ascending aortic width of 7 cm, the Button–Bentall procedure was initially performed and then the CoA was corrected with a second attempt 1 month later. Other patients with cardiac valve pathologies were conservatively followed, because there was no significant stenosis or failure. A patient underwent repair of the ventricular septal defect and atrial septal defect 2 years after the surgery, whereas another patient underwent subaortic discrete membrane resection 1 year later.

During follow-up, a gradient of ≥20 mmHg was accepted as significant on TTE. In addition, because recoarctation (gradient 61 mmHg) was detected 2 months later in a 3-month-old patient who underwent resection and end-to-end anastomosis, balloon dilatation was performed. In

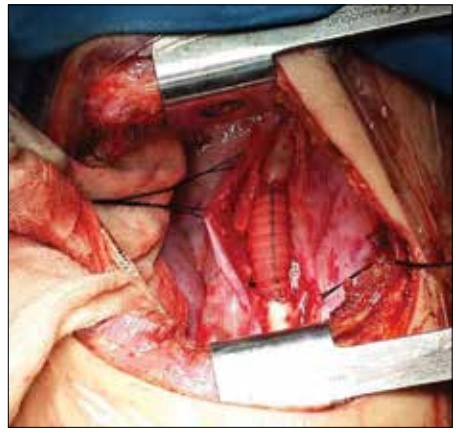

Figure 1. Patch aortoplasty with Dacron graft

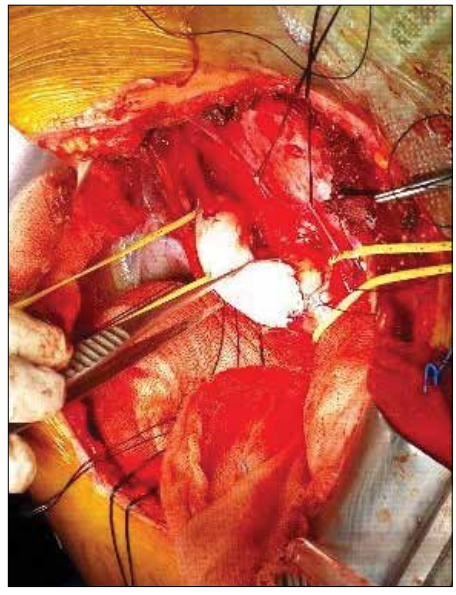

Figure 2. Bypass technique of the lateral isthmus with a PTFE tubular graft

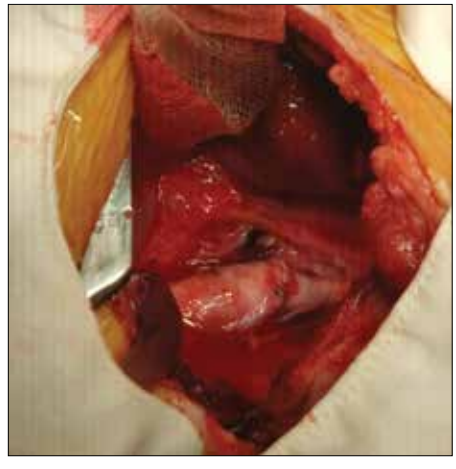

Figure 3. Resection and end-to-end anastomosis

Table 2. Operative details

	n (%)	Mean±SD
Operative techniques		
Resection and end-to-end anastomosis	13 (50)	
Bypass tube graft	6 (23.1)	
Interposition graft replacement	0	
Patch plasty	7 (26.9)	
Operative adjuncts		
Cross-clamping time (min)		23.9±10.7
Side clamping time (min)		19.8±8.6
Left heart bypass	0	
Proximal to distal shunt	1 (3.8)	
Cerebrospinal fluid drain	1 (3.8)	
Concomitant procedures		
PDA ligation/division	5 (19.2)	
Aberrant RSA reimplantation	1 (3.8)	
Subclavian artery reimplantation	2 (7.7)	

PDA: patent ductus arteriosus; RSA: right subclavian artery; SD: standard deviation

Table 3. Postoperative early- and late-stage outcomes

	n (%)
Recurrent laryngeal injury	2 (7.7)
Spinal cord injury	0
Re-intervention for bleeding	1 (3.8)
Renal insufficiency	0
Respiratory (Re-intubation)	2 (7.7)
Chylotorax	2 (7.7)
Pneumothorax	3 (11.5)
İnfection	
Wound	1 (3.8)
Prosthesis	0
Hospital stay (days)	8.4±4.6
Mortality	
Early	1 (3.8)
Late	1 (3.8)
Recoarctation	2 (7.7)
Postoperative aneurysm/pseudoaneurysm	0

Table 4. Late-stage results of different surgical technique

	Resection and end-to-end anastomosis (n=11)			Patch plasty/graft interposition (n=15a)		
	Mean±SD		p value	Mean±SD		p value
	Preoperative	postoperative		Preoperative	postoperative	
Gradient	60.9±15.3	30.7±17	0.001	53.9±24.6	18.3±7.2	0.002
LVEDD	24.8±9.1	22.8±7.8	0.56	48±12.6	36.8±7.8	0.041
LVESD	15.3±5.9	14±6.9	0.64	30.9±9.6	22.4±5.1	0.08
EF	65.9±6.3	69.5±7.9	0.24	65.6±9.1	68.8±7	0.44

a: Data are not available for two patients
LVEDD: left ventricular end diastolic diameter; LVESD: left ventricular end systolic diameter; EF: ejection fraction; SD: standard deviation

a 19-year-old patient who underwent patch plasty, recoarctation was detected at 3 years (gradient 30 mmHg) and balloon dilatation and stenting were performed. Postoperative early- and late-stage outcomes and complications of the patients are shown in Table 3.

In patients who underwent resection and end-to-end anastomosis based on postoperative TTE results during follow-up, a lower statistically significantly gradient compared with the preoperative period (p=0.001) was noted. Despite the decrease in the left ventricular systolic diameter (LVSD) and the increase in the ejection fraction (EF), it did not reach statistical significance. In patients who underwent patch plasty or graft interposition, the low values of gradient and left ventricular diastolic diameter in the postoperative follow-up were statistically significant (p=0.002, p=0.041, respectively). However, the decrease in the LVSD and increase in the EF were not statistically significant compared with the baseline values (Table 4).

Discussion

CoA in adolescents and adults is often complicated with the occurrence of aortic aneurysms, dissections, heart valve disease, and other cardiovascular diseases. Earlier, CoA was evaluated as the localization anomaly of the aorta; however, it is currently considered as part of a broad-spectrum pathology. The main goal of surgical treatment in CoA is the removal of stenosis. The surgical technique is selected according to the length of the coarcted segment, localization with the ductus, status of the collateral circulation in the distal aorta, and atherosclerotic alterations in the aortic wall [1, 2]. Resection and direct end-to-end anastomosis or subclavian flap arterioplasty are the most commonly used techniques for the treatment of CoA in the infantile period because anatomic conditions are more favorable. Subclavian flap arterioplasty and patch graft aortoplasty have been developed as an alternative to resection and direct end-to-end anastomosis in which more than one-half of patients experience late-onset recoarctation problems [1-3]. Extended resection and end-to-end anastomosis are the other techniques to reduce the incidence of recoarctation in the infantile period [1]. Novel suture materials have been developed in recent years, and resection at an early age and end-to-end anastomosis technique have been shown to reduce the rate of recoarctation in patients with CoA.

In the present study, resection and end-to-end anastomosis were performed in all of the patients in the infantile period. In two patients, posterior coarctation membrane was also resected. In a three-month-old patient who underwent resection and direct end-to-end anastomosis, recoarctation developed and balloon dilatation was performed.

In 1961, Vosschulte was the first to describe the prosthetic patch graft aortoplasty technique

[2]. Using this technique, the ductal tissue was ligated or divided and then a longitudinal incision was made through the coarctation segment, and the prosthetic material was placed on the expanded stenotic region. This technique can also be applied to longer segments of coarctation. The circular suturing technique which may cause recoarctation was not used, and aortic mobilization and ligation of the intercostal arteries were less required. In literature, compared with resection and end-to-end anastomosis techniques, aortic aneurysms were reported in a range of 18%–51% in the long term, although the recoarctation rates were between 5% and 12% [3]. Although the use of a more distensible PTFE patch instead of a Dacron® graft was initially promising, it has been shown that there is a risk of aortic aneurysms and recoarctation at a rate of 7% and 25%, respectively.

Furthermore, some authors have suggested that the posterior wall membrane resection plays an important role in the development of aneurysms in patients undergoing patch graft aortoplasty and, therefore, the posterior membrane should not be resected [4, 5]. In our study, patch graft aortoplasty was performed in seven adult patients. The development of aneurysms, pseudoaneurysms, and associated aortobronchial fistulas following the repair of CoA through patch graft aortoplasty is rare and these can be considered as late complications [6]. In the present study, none of the patients who underwent patch aortoplasty experienced such complications in the late period.

As a distinct technique, subclavian flap aortoplasty technique involves no circular suture line and the use of graft material is not required. This technique can be used to repair long-segment coarctations. However, subclavian steal phenomena may develop with this technique, as the left subclavian artery is ligated.

The resection and graft interposition was first described by Gross in 1951 [3]. This technique is not suitable for pediatric patients, because it restricts the development of the aorta. However, bypass grafting is an appropriate technique particularly for patients with aneurysms, long-segment coarctation or post-recovery aneurysms, and adult patients with diffuse collateral circulation and coarctations. Therefore, artificial bypass grafting was preferred in these patients to prevent complications (i.e., spinal cord complications, bleeding, and aneurysm development) during and after surgery [7]. Artificial bypass grafting can be performed by various approaches such as Lateroisthmic bypass (subclavian artery-descending aorta, aortic arch, or descending aorta-descending aorta), ascending aorta-descending aorta bypass, ascending aorta-abdominal bypass, or axillofemoral bypass [8-10]. Six of our patients underwent artificial bypass grafting, whereas tubular graft interposition was not performed in any patient. None of the patients treated with this method developed graft thrombosis, graft infection, or late-onset aneurysms.

If the duration of the ischemia is long enough or there are major collaterals in the area where the aortic cross-clamp is performed, a left cardiac shunt or shunt between the proximal and distal aorta may be used. In our study, a shunt was used in one patient, whereas cerebrospinal fluid drainage was performed in another patient.

Several studies have shown that balloon angioplasty is a relatively effective procedure in the native coarctation. In literature, the recoarctation rate was found to be between 8% and 32%, and the aneurysm formation rate was 24%. The treatment of recurrent coarctations in children can be achieved by 80% to 93% [11-14]. Eight of 11 patients who were treated during the infantile period in our series underwent preoperative balloon dilatation. In one patient, balloon dilatation was performed after the development of postoperative recoarctation.

The use of endovascular stents in coarctation was first reported in 1991 [15]. The stent provides structural support for the aortic wall; therefore, only vascular dilatation may result in less vessel wall injuries and restenosis. The overall rate of complications was 14.3%, whereas the rate of complication related to aortic wall (i.e., aneurysms, intimal tears, or dissections) was 3.9% [16]. Patients who underwent endovascular stenting in our series were also excluded from the study.

In patients with CoA, the optimal operating time is considered as the infantile period or early childhood. Blood pressure control after surgery is one of the problems to be addressed in all patients with coarctation [17]. Cardiac and vascular damage resulting from long-term exposure to hypertension due to coarctation also plays a major role in the postoperative in-hospital mortality, particularly in adult coarctations. In our study, we found a decreased LVSD and increased EF after CoA repair.

The study has limitations. Ours is a single-center study with a small number of patients; this small number of patients may reduce its statistical power. We hope to have more accurate results with studies involving more number of patients. Our follow-up period was also relatively short.

In conclusion, various surgical techniques have provided the possibility of treatment with very low mortality and morbidity rates even in very young children. Our clinical experience suggests that resection and end-to-end anastomosis is a more appropriate treatment option during infancy, whereas patch plasty or bypass grafting may be preferred in advanced ages (adult patients). However, it should not be assumed that patients undergoing CoA repair are completely treated. Therefore, these patients should be followed-up on a regular basis for late complications.

Ethics Committee Approval: Ethics committee approval was received for this study from the Ataturk University School of Medicine Ethics Committee (Decision Date: 05.05.2017/Decision No: 2/18)

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - U.K., A.C.; Design - U.K., A.C.; Supervision - U.K., A.C.; Resources - U.K., N.B.; Materials - U.K., M.C.; Data Collection and/or Processing - U.K., A.C.; Analysis and/or Interpretation - M.C., N.B.; Literature Search - U.K., A.C.; Writing Manuscript - U.K., A.C.; Critical Review - M.C., H.K.

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