Scimitar Syndrome; Right Time to Operate and Mid-Term Results

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ABSTRACT

Introduction: Scimitar syndrome is a combination of rare congenital cardiopulmonary anomalies that can occur in 3% to 6% of patients with a partial abnormal venous connection. The presence of accompanying cardiac anomalies in these patients and in cases such as severe hypoplasia of the right lung or accompanying pulmonary artery hypertension necessitate early surgery in early infancy.

Patients and Methods: Nine patients with scimitar syndrome operated on in our pediatric cardiac surgery clinic from 2012 to 2020 were retrospectively examined in our study. The ages of the patients ranged from 1 to 47 years, with a mean of 18.1 ± 14.4. Patients' weight values ranged from 9 to 88, with a mean of 47.0 ± 31.5. One patient died and mortality was 11.1%. Of the patients, 4 (44.4%) were male and 5 (55.6%) were female. Patients' pulmonary arterial pressure ranged from 0.15 to 94 mmHg, with a mean of 39.2 ± 22.5.

Results: Close to 25% scimitar vein stenosis or scimitar vein drainage occlusion has been reported in the post-operative period, mostly in the newborn group in the literature. Two patients had non-critical stenosis during the third year follow-up despite the absence of stenosis or occlusion during the first two years of follow-up of nine patients we followed. Their surgical follow-up is still ongoing since they are asymptomatic.

Conclusion: The course of the disease depends on the follow-up of the patient, the timing of the surgery, and the quality of the anastomosis. The follow-up and treatment of these patients will be more accurate in advanced centers experienced in scimitar surgery.

Key Words: Abnormal pulmonary venous connection; congenital cardiopulmonary anomalies; increased pulmonary blood flow; pulmonary hypertension; scimitar syndrome.

Scimitar Sendromu; Ameliyat Zamanlaması ve Orta Dönem Sonuçlar

ÖZ

Giriş: Scimitar sendromu, parsiyel anormal venöz bağlantısı olan hastaların %3 ila %6'sında görülebilen nadir konjenital kardiyopulmoner anomaliler birlikteliğidir. Bu hastalarda eşlik eden kardiyak anomalilerin varlığı ve sağ akciğer ağır hipoplazisi ya da eşlik eden pulmoner arter hipertansiyonu gibi durumlarda erken bebeklik döneminde erken cerrahiyi zorunlu kılar.

Hastalar ve Yöntem: Bu çalışmada, 2012-2020 yılları arasında pediatrik kalp cerrahisi kliniğinde opere edilen dokuz scimitar sendromlu hasta retrospektif olarak incelenmiştir. Hastaların yaşı 1-47 yıl arasında değişmekte olup ortalama yaş 18.1 ± 14.4 yıl olarak bulunmuştur. Hastaların kilo değeri 9-88 kg arasında değişmekte olup ortalama 47.0 ± 31.5 kg olarak bulunmuştur. Bir hasta eksitus olup mortalite oranı %11.1'dir. Hastaların 4 (%44.4)'ü erkek, 5 (%55.6)'i kadındır. Hastaların pulmoner arter basıncı 15-94 mmHg arasında değişmekte olup ortalama 39.2 ± 22.5 mmHg olarak bulunmuştur.

Bulgular: Literatürde postoperatif dönemde daha çoğu da yenidoğan grubunda olmak üzere %25'e yakın scimitar ven stenozu ya da scimitar ven drenajı oklüzyonu bildirilmiştir. Takip edilen dokuz hastanın ilk iki yıllık takiplerinde stenoz ya da oklüzyon görülmemesine rağmen, iki hasta da üçüncü yıl takiplerinde kritik olmayan stenoz görülmüştür. Asemptomatik oldukları için cerrahi takiplerine devam edilmektedir.

Sonuç: Scimitarda hastalığın seyri hastanın takibine, cerrahinin zamanlamasına ve anastomozun kalitesine bağlıdır. Bu hastaların scimitar cerrahisi konusunda tecrübeli ileri düzey merkezlerde takip ve tedavisi daha doğru olacaktır.

Anahtar Kelimeler: Anormal pulmoner venöz dönüş; konjenital kardiyopulmoner anomali; pulmoner hipertansiyon; scimitar sendromu.

INTRODUCTION

Scimitar syndrome is a combination of rare congenital cardiopulmonary anomalies that can occur in 3% to 6% of patients with a partial abnormal venous connection. It is named scimitar because it is characterized by a Turkish Sword (scimitar)-like shadow extending

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towards the cardiophrenic angle in the middle zone of the right lung (Figure 1). The opening of some or all of the right pulmonary veins to the inferior vena cava from below or above the diaphragm by abnormal venous drainage is characterized by varying degrees of severity of right lung hypoplasia (Figure 2). The pulmonary vein or veins of the lower and sometimes middle lobe of the right lung, or rarely the entire right lung, are opened to the upper part of the inferior vena cava, which can be opened to the upper or lower part of the diaphragm in these patients. The lower lobe of the right lung is most commonly hypoplasic and draws its blood from systemic arteries, especially the thoracic or abdominal aorta, rather than the pulmonary artery[1-3].

Symptoms may vary depending on other accompanying anomalies in this patient group. Many patients may remain asymptomatic until adolescence or advanced age or progress to this age with mild to moderate symptoms. In another patient group, the presence of accompanying cardiac anomalies and in cases such as severe hypoplasia of the right lung or accompanying pulmonary artery hypertension necessitate early surgery in early infancy[4].

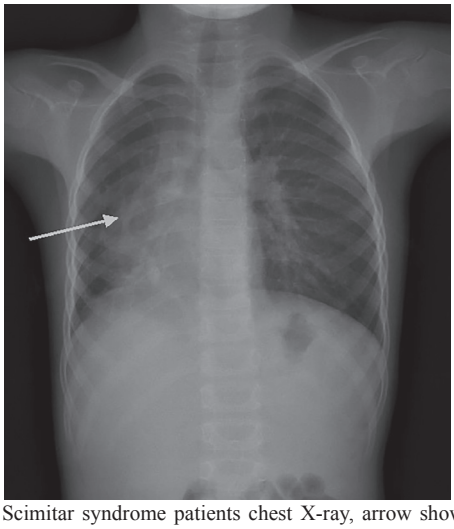

Figure 1. Scimitar syndrome patients chest X-ray, arrow shows scimitar sign is pathognomonic for scimitar syndrome.

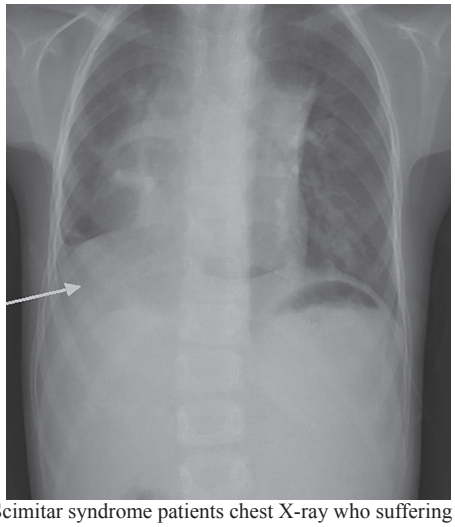

Figure 2. Scimitar syndrome patients chest X-ray who suffering right inferior hypoplasic lung segment with shown by arrow.

Approximately 70% of patients with scimitar syndrome have an associated atrial septal defect. Overall, between 19% and 31% of patients with scimitar syndrome have other accompanying cardiac anomalies. It has also been associated with other cardiac malformations such as Tetralogy of Fallot, ventricular septal defect, aortic coarctation, hypoplastic left heart syndrome, total abnormal pulmonary venous connection, patent ductus arteriosus, cortical striatum, bicuspid aortic valve, and subaortic stenosis even though the syndrome is less common[5,6].

Dupuis et al. examined scimitar syndrome under three main headings: infantile form with symptomatic and pulmonary hypertension, the adult form seen in the older group, another form distinguished by being asymptomatic in infancy and accompanied by congenital heart anomalies associated with it[7].

Surgical correction is usually performed in symptomatic patients or in patients with increased pulmonary blood flow and significant enlargement of the right heart structures. Different surgical approaches have been defined over the years according to the surgeon's preference and case characteristics. The method to be applied depends on the choice of the surgeon depending on the anatomical and pathological characteristics of the patient.

Many clinical studies have been conducted to monitor the natural course of the disease. Clinical follow-up and registration are very valuable to better understand the natural course of the disease. Preoperative and postoperative close follow-up and recording make an important contribution to us in terms of analyzing and evaluating the effectiveness of surgical outcomes both in terms of surgical necessity, surgical timing, and postoperative period.

This study, in which we retrospectively examined the records of nine scimitar syndrome patients in our clinic between 2012 and 2020, aims to analyze the patients operated with the diagnosis of scimitar syndrome, our Kosuyolu experience, surgical results, and follow-up results in a relatively close period of time.

PATIENTS and METHODS

Nine patients with scimitar syndrome operated on in our pediatric cardiac surgery clinic from 2012 to 2020 were retrospectively examined in our study. The records of the patients were accessed from the hospital archive and the hospital information processing system.

Median sternotomy was used as an incision in all patients and all patients were operated on under cardiopulmonary bypass. The arterial cannulation of the patients was performed as aortic and venous cannulation as bicaval. Daily radiological chest X-ray and echocardiography evaluations were performed in the postoperative follow-up of the patients.

Our study is a retrospective, observational, single-center case series study. Age, gender, weight, diagnosis, previous operations, procedure performed, length of hospital stay, and biochemistry results of the patients were recorded.

The surgical technique applied to the patients was preferred to anastomose the scimitar vein to the left atrium. The drains of the patients were removed when 0.100 cc/day cc in adults and less than 5 cc/kg in pediatric patients. The ASA was started as postoperative antiaggregant therapy in all patients.

Retrospectively, ethical rules were followed according to the Declaration of Helsinki. Consents of the patient and their parents were obtained before the operation.

Number Cruncher Statistical System (NCSS) 2007 software (Kaysville, Utah, USA) was used for statistical analysis. Descriptive statistical methods (Mean, Standard Deviation, Median, Frequency, Ratio, Minimum, Maximum) were used to evaluate the study data.

RESULTS

The study included 9 patients operated for scimitar syndrome in our clinic between 2012 and 2020. Symptoms of the patients were asymptomatic, respiratory distress, dyspnea exertional, tachycardia. Two of the patients had right lung hypoplasia, advanced pulmonary hypertension (PHT), and heart failure. One patient also presented with recurrent pneumonia. The remaining patients had moderate advanced PHT. It was not accompanied by heart failure.

Right lung hypoplasia was detected in two patients. Reoperation was not performed in the early period due to bleeding diathesis or any reason. Both patients had scimitar vein anastomotic stenosis during the mid-term follow-ups. Follow-up was continued since they were asymptomatic. Two patients underwent catheterization and closure of the abnormal systemic arterial structure by our pediatric cardiology team in the preoperative period (Figure 3, 4).

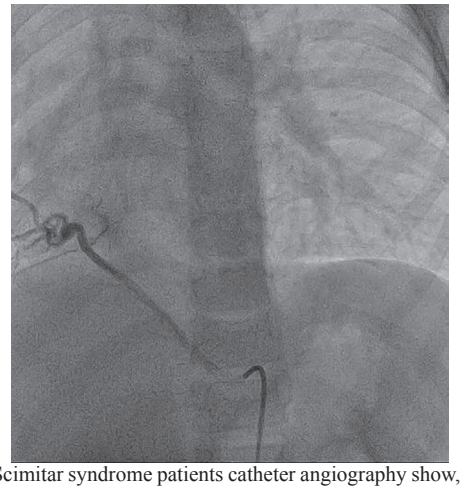

Figure 3. Scimitar syndrome patients catheter angiography show, abnormal systemic arterial feeding in catheterization.

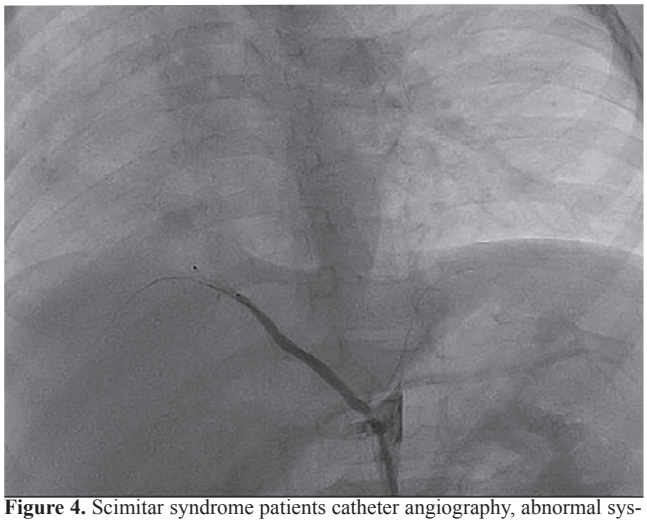

Figure 4. Scimitar syndrome patients catheter angiography, abnormal systemic arterial disconnection with coil embolization by our pediatric cardiology team in the preoperative period

Two patients required prolonged mechanical ventilation with the diagnosis of pneumonia and they were extubated on the postoperative seventh day, in addition to postoperative complications and postcardiac surgery complications associated with scimitar repair. One patient, who was operated on with the symptoms of pulmonary hypoplasia, PHT, and heart failure at 14 months of age, died on the postoperative 20th day with the diagnosis of low cardiac output in the postoperative period.

The ages of the patients ranged from 1 to 47 years, with a mean of 18.1 ± 14.4. Patients' weight values ranged from 9 to 88, with a mean of 47.0 ± 31.5. Of the patients, 4 (44.4%) were male and 5 (55.6%) were female. One patient died and the mortality was 11.1%. Patients' pulmonary arterial pressure ranged from 0.15 to 94 mmHg, with a mean of 39.2 ± 22.5. Patients' cardiopulmonary bypass time ranged from 53 to 103 mmHg, with a mean of 73.9 ± 19.6. Patients' ventilator time

Table 1. Data of patients as a result of descriptive analysis

	N	Minimum	Maximum	Mean	SD
Age	9	1	47	18.1	14.4
Weight	9	9	88	47	31.5
Pulmonary artery pressure	9	15	94	39.2	22.5
Cardiopulmonary bypass time	9	53	103	73.9	19.6
Ventilator time	9	1	20	4	6.1
Length of stay in ICU	9	2	20	5.2	5.7
Length of stay in ward	9	0	9	6.6	2.9
Length of stay	9	8	20	11.8	3.6

ICU: Intensive care unit, SD: Standart deviation.

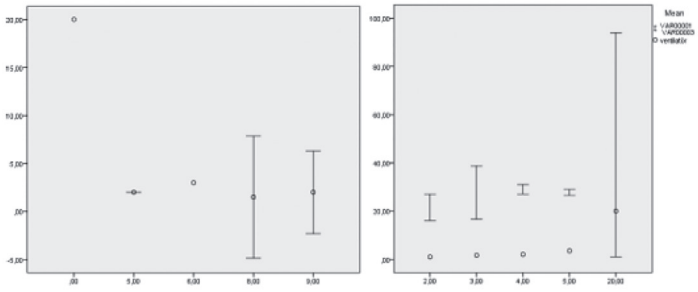

Figure 5. Box plot graph of pulmonary pressures and length of stay in intensive care and in ward.

ranged from 1 to 20 days, with a mean of 4 ± 6.1. Patients' length of stay in intensive care ranged from 2 to 20 days, with a mean of 5.2 ± 5.7. Patients' length of stay in the ward ranged from 0 to 9 days, with a mean of 6.6 ± 2.9. Patients' total length of hospital stay ranged from 8 to 20 days, with a mean of 11.8 ± 3.6 (Table 1, Figure 5).

DISCUSSION

The causes affecting the occurrence of symptoms in scimitar syndrome have a wide range of causes. The presence of other associated congenital heart malformations, the amount of blood spilled into the inferior vena cava, the presence of scimitar vein obstruction, and the degree of symptoms may vary depending on the degree of pulmonary artery flow. The presence of bronchial anomalies in the right lung or lower lobe may also cause secretion, lobar infections, and hemoptysis. There may be difficulties and confusion in diagnosing, especially in children and young adults with congenital heart lesions due to the wide range of symptoms at the onset of the clinic. The diagnosis is usually made by a careful cardiologist during echocardiography follow-up for another reason since these patients are generally asymptomatic[8-12].

Pulmonary hypertension occurs frequently in scimitar syndrome and is often accompanied by congenital heart malformations or abnormal major systemic artery source in the infantile age group. Similarly, this association can be seen in the adult age group. A less common cause of pulmonary arterial hypertension is the presence of pulmonary vein stenosis[13,14].

The most difficult decision in follow-up and treatment in this patient group is probably for patients with asymptomatic or mild symptoms, which are less severe forms. Deciding on follow-up or surgical correction in these patients or deciding the timing of surgery makes it difficult for the patient to make the most accurate decision, as it requires not considering many variables. We organize our surgical indications considering the progressive pulmonary artery pressure and the symptoms of patients in our clinical practice. In the follow-ups, we made a surgical decision considering the symptoms of two patients with high pulmonary artery pressure at the border after three years of follow-up. Three patients who went with pulmonary hypertension had a decrease in postoperative pressures. And the clinical improvement was observed in follow-ups. None of the patients had stenosis or occlusion in the early period. We attribute these positive results to the fact that the majority of patients have completed lung maturation, albeit a little after the neonatal period. However, both patients had scimitar vein anastomotic stenosis during the mid-term follow-ups. We continue echocardiographic follow-up of these asymptomatic patients[15-18].

Patients requiring surgical treatment under one year of age are usually very ill and usually have other accompanying cardiac anomalies. In addition, their lung is hypoplasic and they have relatively high operative mortality. The complication rate in the newborn group is higher in the early and long term compared to elderly patients. The presence of left-to-right shunt in ASD, which is the most common accompanying factor with scimitar syndrome, makes these patients symptomatic earlier and enables them to be diagnosed earlier[19]. The patient, who was operated on due to signs of pulmonary hypertensive (94 mmHg) pneumonia at 14 months of age and heart failure accompanied by pulmonary hypoplasia, died on the postoperative 20th day.

The therapeutic occlusion of abnormal systemic arterial feeding to the right lung provides a clinical improvement, especially in SS patients accompanied by congestive heart failure, as well as a significant contribution to the regression of congestive heart failure signs and postponement of surgical treatment to older ages[20]. Abnormal systemic arterial feeding due to abdominal and thoracic aorta was closed in three patients during interventional catheterization in our clinical follow-ups (Figure 3, 4).

The presence of pulmonary hypertension and associated CHD is a negative prognostic factor on the survival of patients regardless of whether patients are treated surgically[17,18]. Of our 9-person group, 5 had PHT. Two had additional CHD and three had isolated PHT. Vida et al. observed PHT independent of the presence of accompanying CHD in nearly 30% of all scimitar syndrome patients in their study.

Close to 25% scimitar vein stenosis or scimitar vein drainage occlusion has been reported in the postoperative period, mostly in the newborn group in the literature[15,16]. Two patients had

stenosis in the 3rd year despite the absence of this during the first two years of follow-up of nine patients we followed up. Follow-up is continued since the patient is asymptomatic without the need for additional surgery.

Scimitar syndrome alone can cause pulmonary artery overload, resulting in the development of PHT. Surgical treatment is inevitable when pulmonary overload Qp:Qs > 1.5:1[20,21].

It is recommended in the literature to have magnetic resonance imaging (MRI) imaging during follow-ups. The diagnosis was made by echocardiography in our patient group. Scimitar vein anatomy, CHD anomaly in addition to the presence of PHT, imaging of abnormal systemic arterial feeding to the lung, and evaluation of the presence of shunt were also catheterized for diagnosis[19-21].

The systemic arteries feeding the affected lobe(s) should be obstructed and eliminated by reducing blood flow and shunt amount if the presence of a large systemic abnormal arterial feeding to the right lung is detected by preoperative pulmonary excessive circulation. This has often been advocated as the best and simplest treatment that improves clinical symptoms and lowers pulmonary arterial pressure, especially in infants with congestive heart failure[14,15]. Two patients in our patient group underwent catheterization and closure of the abnormal systemic arterial structure by our pediatric cardiology team.

Surgical repair of scimitar syndrome consists of redirecting pulmonary venous drainage to the left atrium. It is either to transmit abnormal drainage to the left atrium via a tunnel (baffling) or to transect "scimitar drainage" near the entrance of the inferior vena cava and then reinserted directly into the left atrium[11-14]. Both baffle repair and direct reimplantation of scimitar vein have been in use for many years. There is no consensus yet about the superiority of one over the other. It is up to the surgeon to choose the appropriate surgical method in the case according to the anatomical and pathological characteristics of the patient. We applied the technique of anastomosing the left atrium by transecting the scimitar vein to all patients in our patient group. We do not yet have sufficient clinical experience since we do not prefer the baffle technique in our clinical practice.

Huddleston et al.[21] reported right lung lobectomy or pneumonectomy (as primary therapy or if there is postoperative stenosis after unsuccessful repair) in symptomatic patients with recurrent pulmonary infections and bleeding even though repair of abnormal venous return and ligation of collaterals are generally recommended. It is an option that leads us to similar results.

Right lung pneumonia is generally well-tolerated in children, especially in patients with affected lung hypoplasia. Lobectomy can only be performed when the abnormal vessel empties that lobe and care must be taken to ensure that the remaining lung function remains adequate. We did not prefer pneumonectomy surgery even though there were patients in our study group with pneumonia and right lower lobe hypoplasia.

It suggests that scimitar syndrome is not a simple and benign congenital heart disease and is usually associated with other heart anomalies that play an important role in long-term outcomes. Most of our patients presented with preoperative symptoms representing one of the main indications for surgical repair, especially in the presence of pulmonary arterial hypertension.

Overall, it has shown that corrective surgery can be performed safely with a low mortality and morbidity rate, regardless of the type of corrective surgery technique used. It should be noted that the presence of PHT is associated with a higher mortality rate. In addition, scimitar vein anastomotic stenosis may occur, which will require re-surgery in the mid-term, regardless of the type of surgery, or which will require follow-up even though it is asymptomatic.

There are sources indicating that the results of surgical treatment are related to the age of the patient. We cannot make this generalization in our study group because our age group was between 1-47 years old and there are a total of nine patients. As a general rule, patients less than 1 year of age in need of treatment are usually very ill and have relatively high operative mortality and complication rate whereas older patients have better outcomes both in the early and long term. Previous reports have shown that delayed surgical treatment, especially in infants with congenital heart lesions, may lead to irreversible pulmonary vascular obstructive disease that may jeopardize the surgical outcomes of these patients[18-21].

The present study had some limitations. We used PA chest radiography, control echocardiography, and improvement in clinical symptoms in our cohort for patients even though there are publications suggesting postoperative right lung evaluation with pulmonary scintigraphy. None of the patients underwent re-catheterization with the suspicion of postoperative stenosis or occlusion. However, follow-up with scintigraphy may still provide clearer information for postoperative right lung anatomy and especially for monitoring its functional status.

CONCLUSION

We have reported our Kosuyolu experience. Long-term follow-up of these patients, which includes years, does not make it possible in the conditions of Turkey due to both healthcare system and social reasons even though our preoperative and postoperative follow-ups are regularly performed by the pediatric cardiology and surgery team with councils.

Surgical treatment in these patients is useful in reducing symptoms, even in patients with CHD and pulmonary hypertension, as well as asymptomatic or mildly moderate

symptoms. However, the most feared complication of surgery is occlusion or stenosis in the middle and late periods. The incidence of this complication has increased up to 25% in some publications[16-21]. The most important cause of symptoms is thought to be due to increased pulmonary blood flow. Increased pulmonary overload to the scimitar vein is present.

Two opinions have been presented in the literature regarding strategies to increase the overall survival of the patients. There are opinions that correction of cardiac anomaly accompanying the same session at an early age and surgery with therapeutic occlusion of abnormal systemic arterial feeding to the lung (performed after) may be beneficial in selected patients. Many sources argued that follow-up with MRI in patients with asymptomatic or mild symptoms and surgical correction in advanced ages are more useful because they complete lung maturation. Thus, they argued that the follow-up of scimitar syndrome to advanced ages minimized the risk of late morbidities such as postoperative stenosis and occlusion.

As a result, the course of the disease depends on the follow-up of the patient, the timing of the surgery, and the quality of the anastomosis. The follow-up and treatment of these patients will be more accurate in appropriate centers.

Ethics Committee Approval: The study was approved by the Kartal Koşuyolu High Specialization Training and Research Hospital Clinical Research Ethics Committee (Date: 13.07.2021, Decision No: 2021/7/505).

Informed Consent: Informed consent was obtained.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept/Design - KK; Analysis/Interpretation - KK; Data Collection - EA; Writing - EA, KK; Critical Revision - HC; Statistical Analysis - FT; Overall Responsibility - HC; Final Approval - All of authors.

Conflict of Interest: The authors have no conflicts of interest to declare

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