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Factors affecting complications in 31 cases of elastofibroma dorsi after marginal resection in a single center

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ABSTRACT

Aim: Elastofibroma dorsi (ED) is an uncommon benign connective-tissue tumor, usually seen in the subscapular region of women after the fifth decade. We present the clinical features, management, and long-term outcomes of cases of ED treated surgically in a single institution.

Material and Method: The data of 31 patients (7 male, 24 female) with a histopathological diagnosis of ED between January 2010 and January 2021 and mean age of 56.6 years were reviewed retrospectively from their records. The mean follow-up duration was 80.2 (19-144) months. Nine cases were bilateral. Marginal resection surgery was performed in all cases diagnosed radiologically and clinically, and preoperative biopsies were performed for three patients. The results were evaluated using a visual analogue scale (VAS) for pain during follow-up.

Results: Complications such as chronic pain (n=5), hematoma (n=5), seroma (n=5), and infection (n=2) were seen in 11 patients (35%) in the early postoperative period and improved over the course of follow-up. A local recurrence observed in one patient during follow-up was re-excised. Significantly more complications were observed in patients with bilateral ED (p=0.015), manual laborers and heavy laborers (p=0.013), patients with comorbidities (p=0.006), those who slept in the supine position (p=0.031), and those who underwent synchronized surgery (p=0.013). In addition, statistically significantly more complications were observed in cases of masses with longer longitudinal length (p=0.016), patients with longer preoperative symptom duration (p=0.009), and longer operative times (p=0.025). The average VAS score improved significantly from 4.97 to 1.52 after surgery (p<0.001).

Conclusion: While satisfactory results were obtained over a long follow-up duration after marginal resection, many complications were encountered in the early postoperative period. The decision for resection should be made after a comprehensive evaluation of symptoms and lesions, patients should be informed about complications, and follow-up periods should be prolonged after meticulous surgery.

Keywords: Elastofibroma dorsi, marginal resection, subscapular mass, benign tumor

INTRODUCTION

Elastofibroma dorsi (ED) is a slow-growing benign fibroblastic pseudotumor in the infrascapular region. ED typically presents as an ill-defined mass lying over the costal periosteum at the 6th to 8th ribs, underneath the rhomboid major, latissimus dorsi, or serratus anterior muscles (1,2). It is especially common in women over the age of 50 and it constitutes 1-2% of chest wall primary tumors without malignant potential (1,3). However, it has been stated in some studies that its incidence varies between 1% and 16% with asymptomatic cases (3,4). It usually entails a unilateral lesion (60% right-sided), but it has been reported to be bilateral in 10-66% of cases (3-5).

Most patients are asymptomatic. With increased size of the mass, patients present with complaints of local pain that increases with shoulder movements, limitation of movement, a mobile mass with a rubbery consistency, or scapular snapping (5,6). Laboratory and pulmonary function test results are normal (2). Because of its typical localization and clinical features, it is often diagnosed after radiological examinations. Magnetic resonance imaging (MRI) is the mainstay of diagnosis of ED due to its high sensitivity and specificity (7). MRI demonstrates a heterogeneous, unencapsulated, soft tissue mass that is lenticular, poorly circumscribed, and most often with intermixed linear-intensity adipose tissue within fibrous tissue (1,7) (**Figure 1**).

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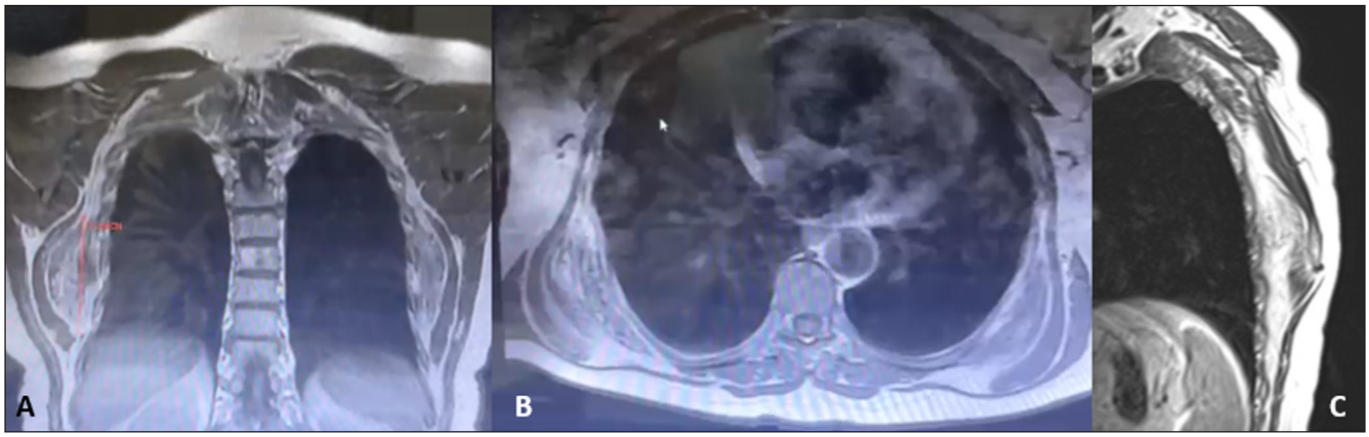

Figure 1. Coronal (A), axial (B), and sagittal (C) MRI images of bilateral elastofibroma dorsi.

Although it has been stated that biopsy is often unnecessary, it is recommended in suspicious cases with atypical clinical and radiological findings without a contralateral mass (1,8). It can be confused with other tumors of the periscapular region and misdiagnosed as sarcoma. The definitive diagnosis is made histopathologically (1). Marginal resection with a muscle-sparing approach is a generally accepted technique for symptomatic and/or large ED (1,4,5). Although significant improvement in the preoperative symptoms of patients is observed, postoperative complications such as seroma and hematoma are seen at varying rates due to the poorly circumscribed mass and its high vascularity (4,9,10).

As a limited number of studies have reported the long-term surgical outcomes of ED and the number of cases is small, clearly defined treatment guidelines and optimal approaches have not yet been determined. In this study, we aimed to determine the clinical and demographic factors affecting the postoperative complications and long-term results of patients with ED in a single center and to contribute to the literature by sharing our experiences.

MATERIAL AND METHOD

The study was carried out with the permission of University of Health Sciences Dr. Abdurrahman Yurtaslan Ankara Oncology Health Applications and Research Center Clinical Researches Ethics Committee (Date: 06.10.2021, Decision No: 10-1419). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

Forty masses of 31 consecutive patients (7 male and 24 female), who were surgically treated by the orthopedics and traumatology, and thoracic surgery clinics in our oncology center between January 2010 and January 2022 and were diagnosed with ED histopathologically with a follow-up period of at least 12 months, were included in this study. Mean age was 56.6±12.06 (19-74) years. Demographic data, clinical presentations, and complications were obtained from the patients' records and analyzed retrospectively (**Table 1**).

Table 1. Perioperative demographic and clinical features and complications of 31 patients with elastofibroma dorsi

Parameters	n (%) or mean±SD
Age, mean±SD	56.68±12.06
Gender, n (%)	
Female	24 (77.4%)
Male	7 (22.6%)
Occupation, n (%)	
Housewife	16 (51.6%)
Civil officer	6 (19.4%)
Manual/heavy labor	9 (29%)
BMI, mean±SD	26.56±3.82
BMI <25	11 (35.5%)
BMI ≥25	20 (64.5%)
Comorbidities, n (%)	13 (41.9%)
Operation side, n (%)	
Right	14 (45.2%)
Left	8 (25.8%)
Bilateral	9 (29.0%)
Surgery, n (%)	
Unilateral	22 (71.0%)
Synchronized bilateral	6 (19.4%)
Two-sequence bilateral	3 (9.7%)
Time of follow-up, weeks	
Mean±SD	80.26±33.06
Longitudinal length of ED	
Mean±SD	70.55±19.97
Symptoms, n (%)	
Pain	21 (42%)
Feeling of mass	15 (30%)
Restriction of movement	8 (16%)
Snapping scapula	6 (12%)
Duration of symptoms, months	7.61±6.83
Sleeping in the supine position, n (%)	
Yes	14 (45.2%)
No	17 (54.8%)
VAS score	
Preoperative	4.97±1.27
Postoperative	1.52±1.06
Drain removal time, days	
Mean±SD	1.65±0.87
Complications, n (%)	
None	20 (64.5%)
Pain	5 (16.1%)
Seroma	5 (16.1%)
Hematoma	5 (16.1%)
Infection	2 (6.4%)
Recurrence of ED, n (%)	1 (3.2%)
Operative time, minutes	53.55±4.20
Hospitalization time, days	
Mean±SD	2.55±0.40

SD: Standard deviation, BMI: Body mass index, ED: Elastofibroma dorsi, VAS: Visual analogue scale

The masses in our patients were located on the right side (n=14), left side (n=8), and bilaterally (n=9). Twenty-five patients (7 manual laborers, 2 heavy laborers, and 16 housewives) had jobs that required more than 10 years of extensive physical effort. All of these cases were symptomatic; the most common symptoms were pain and palpable mass. The overall mean of the longitudinal length of excised masses was 70.55±19.97 (45-115) mm. All of our patients had subscapular masses, and plain conventional X-rays and MRI (**Figure 1**) were requested as radiological examinations. The diagnosis was made based on physical examination, characteristic findings, and radiological imaging. Biopsy was performed for 3 atypical cases with difficulty in diagnosis.

Atypical and prominent symptoms (27/31) and patient's preference (4/31) were considered as surgical indications. The average time from the onset of symptoms to surgery was 7.61±6.83 (2-36) months. After general anesthesia, marginal resection surgery with a muscle-sparing approach was performed with an oblique incision from the lower end of the scapula (**Figure 2**). While the mass was bluntly separated from the upper muscle planes, it was dissected from the rib cage and scapula with the help of cautery. After meticulous hemostasis, a Hemovac drain was placed in the operation area for large or difficult-to-dissect tumors. Penrose drains were used in other cases. The mean hospitalization time was 2.55±0.40 (1-9) days and the time to drain removal was 1.65±0.87 (1-4) days. No patients required blood transfusions. Synchronized bilateral surgery was performed in 6 of 9 bilateral cases, while surgery was performed in two consecutive sessions for the remaining 3 patients.

The diagnosis was confirmed histopathologically. Local cryotherapy for the first 48 hours postoperatively and immobilization for 1 week was recommended. A shoulder arm sling and compression bandage were applied for the first 15 days. Passive shoulder movements were started at the end of the 1st week and active movements from the end of the 1st month. In the postoperative period, no restrictions were applied to movements, except for shoulder abduction and heavy activities of the affected upper extremity. Clinical outcomes were evaluated preoperatively and postoperatively using a visual analogue scale (VAS) for pain. All of the complications that we observed in the postoperative period improved during follow-up. In the follow-ups, it was observed that our patients had returned to their normal daily living activities with almost complete shoulder movement without pain.

Statistical analysis: All analyses were carried out using IBM SPSS Statistics 25.0 (IBM Corp., USA). Means and standard deviations were used to represent quantitative data. A statistical significance threshold of $p<0.05$ was applied. Preoperative and postoperative VAS scores were compared using the paired-sample t-test. The Mann-Whitney U test was carried out to compare variables without normal distribution. Comparisons of categorical data between groups of dependent variables were performed with chi-square tests. Pearson chi-square, Fisher exact, or continuity correction was applied as appropriate. Binary logistic regression analysis was performed to evaluate variables that could be associated with whether or not complications occurred. Multivariate logistic regression analysis was used to determine the factors associated with complication and recurrence rates.

RESULTS

A total of 18 complications were observed in 11 (35.4%) patients during an average follow-up duration of 80.2 (19-144) months after histologic resections of masses with tumor-free margins. Specific complications were as follows: pain (n=5), hematoma (n=5), seroma (n=5), and infection (n=2). Complications were managed conservatively (needle aspiration, puncture, compressive dressing, antibiotherapy, immobilization) in 10 of 11 cases. One patient was re-operated on in the first postoperative month for wound infection. While the mean VAS score was 4.9 (3-7) preoperatively, it decreased to 1.8 (0-5) postoperatively, which constituted a statistically significant difference ($p<0.001$) (**Table 2**). All our patients verbally stated that all complaints were resolved and limitations of shoulder movements improved after marginal resection.

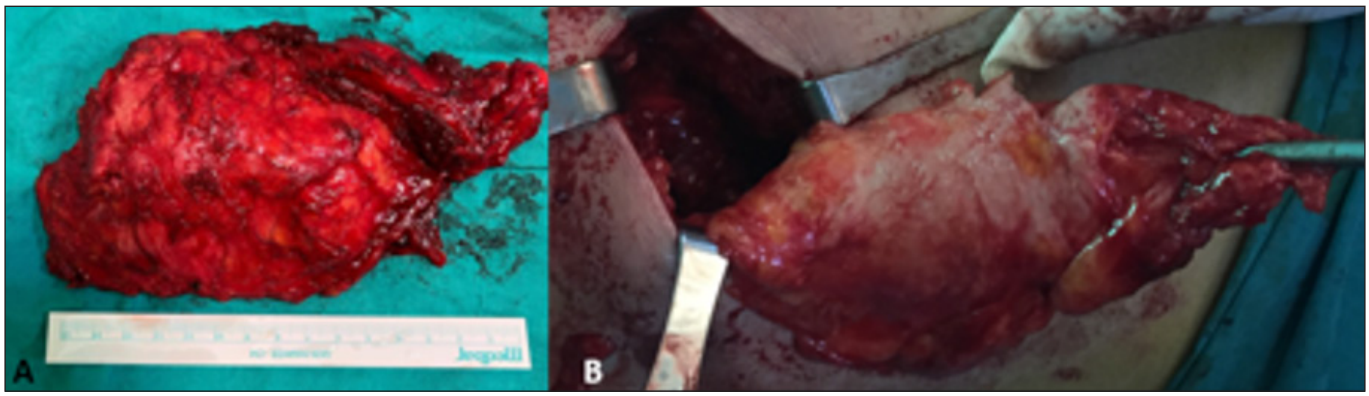

Figure 2. Post-excision (A) and intraoperative (B) images of elastofibroma dorsi mass

Table 2. Comparison of preoperative and postoperative visual analogue scale scores

	Mean	Standard Deviation	Std. Error of Mean	t	df	p
Preoperative and postoperative visual analogue scale scores	3.452	0.768	0.138	25.035	30	<0.001*

* Paired samples t-test

The complication rates were significantly higher among patients with bilateral ED, those with comorbidities, and those who were manual or heavy laborers (p=0.015, p=0.006, and p=0.013, respectively). The complication rate was also higher among patients who underwent synchronized surgery (n=6) compared to unilateral surgery (n=22) and among patients who slept in the supine position (n=14) (p=0.013 and p=0.031, respectively). Delayed wound healing, hematoma, and seroma were more common among patients who slept in the supine position in the group with complications, being seen in 8 of 11 cases. There was no difference in complication rates in terms of age, gender, or body mass index (BMI) (p>0.05) (**Table 3**). Multivariate logistic regression analysis showed that longitudinal mass length, operative time, and preoperative symptoms were positively correlated with complications (p=0.016, p=0.025, and p<0.01, respectively). While a significant positive relationship was found between drain removal time and complications, the positive relationship between hospitalization time and complications was not statistically significant (p=0.041 and p=0.068, respectively).

Table 3. Analysis of risk factors for postoperative complications and recurrence

Factors	Complications p-value*	Recurrence p-value*
Age	0.157	0.242
Gender (male/female)	0.652	1.000
Unilateral or bilateral mass	0.015	0.503
Occupation (housewife, manual labor, heavy labor)	0.013	0.118
BMI <25 or BMI ≥25	0.262	1.000
Comorbidities (yes/no)	0.006	0.196
Sleeping in the supine position	0.031	1.000
Preoperative biopsy diagnosis (yes/no)	0.121	1.000
Unilateral/synchronized surgery	0.013	1.000

*: Chi-square test, BMI: Body mass index, ED: Elastofibroma dorsi

The longitudinal length of the masses was positively correlated with the weight of patients, BMI, and postoperative VAS scores (p=0.013, p=0.06, and p=0.037, respectively). Length was not correlated with preoperative VAS score, duration of symptoms, operative time, or hospitalization time (p>0.05) (**Table 4**). There were no significant differences in the parameters evaluated in terms of recurrence (p>0.05) (**Table 3**). However, the local recurrence observed in one patient in the bilateral surgery and manual/heavy labor groups, who slept in the supine position and had a high preoperative VAS score, was re-excised.

Table 4. Multivariate logistic regression analysis of factors associated with complications

Model	Unstandardized Coefficients		Standardized Coefficients	t	Sig.
	B	Std. Error	Beta		
(Constant)	-1.265	0.445		-2.841	0.010
Age	0.003	0.004	0.067	0.734	0.471
BMI	0.019	0.014	0.156	1.364	0.187
Time of follow-up	-0.001	0.002	-0.088	-0.808	0.428
Longitudinal length of the mass	0.006	0.002	0.268	2.615	0.016
Duration of symptoms	0.020	0.007	0.283	2.875	0.009
Preoperative VAS score	-0.061	0.051	-0.164	-1.182	0.250
Duration of operation	0.006	0.003	0.297	2.412	0.025
Time to removal of drain	0.145	0.091	0.268	1.586	0.128
Duration of hospitalization	0.065	0.032	0.312	2.062	0.052

DISCUSSION

ED is an uncommon, benign, poorly circumscribed connective-tissue tumor classically located in the subscapular region (5,8). Although ED is mostly unilateral and usually located on the right side, some studies reported bilateral involvement in 10% to 50% of cases (2,4,5,11,12). The rates of right-sidedness (45.2%), left-sidedness (25.8%), and bilaterality (29.0%) in our study are consistent with the literature. In addition, it has been reported that large lesions are more common in women and the elderly (3,5,11). In our study, which had a female/male ratio of 24/7, no significant relationship was found between the size of the masses and gender or age. This is likely because our study group comprised symptomatic patients who all underwent surgery.

ED is often asymptomatic, and symptoms vary depending on the size and localization of the lesion (5,6). In a series of 76 cases that were treated surgically, the most common symptoms were pain and functional limitations (9), while in our series, pain (42%) and feeling the mass (30%) were most common. Limitations of shoulder abduction and feelings of stiffness were particularly observed in symptomatic patients in previous reports (5,6), consistent with our study, and these symptoms improved after surgery. While some researchers reported a relationship between the dimensions of the ED and the severity and presence of symptoms (12), other findings have suggested no such relationships (11). Our study revealed a positive and significant correlation between tumor size and variables including weight, BMI, postoperative VAS score, and complications. The absence of a relationship between tumor size and

symptoms in our study was probably due to the fact that our study group included only symptomatic patients who underwent surgery.

Although different theories have been proposed for the etiology of ED, the main accepted view is elastin degeneration caused by repetitive microtraumas (2,12-14). It is frequently seen in people who perform manual labor involving arm strength (15) and in housewives (5), and it has also been reported 10 years after latissimus dorsi flap harvesting (16). In our study, 25/31 patients (Housewife-16, Manual/heavy labor-9) did work that required manual labor and more complications were seen in this group; these findings support the microtrauma hypothesis.

If all pathognomonic criteria (age, localization, and snapping sound) are present, the lesion can be confidentially diagnosed with at least one radiological examination, preferably MRI (7,8,14). It has been shown that the diagnostic value of radiological imaging for ED is high and biopsy is not necessary for clinically and radiologically typical cases (1,7,17,18). Since we preferred MRI for diagnostic purposes, we performed a biopsy for only one patient; two patients admitted to our clinic had biopsies performed at another hospital previously.

As in our cases, no metastasis or malignant transformation has been reported in the literature despite the proximity of ED to the thorax and its high vascularity (7). The differential diagnosis should consider benign tumors such as fibrolipoma and neurofibroma, aggressive tumors such as desmoid fibromatosis, and sarcomas (7,11,12). Due to the variety of clinical manifestations, ED may be misdiagnosed as rotator cuff tear, subacromial bursitis, cervical pathologies, or chronic back pain. In order not to cause delays in diagnosis, shoulder and neck problems should be revealed with a careful physical examination (5,9,14). The fact that our patients were treated in different clinics during the preoperative symptom duration of 10 months supports this.

The management and follow-up of ED are somewhat controversial. The standard surgical treatment method recommended for ED is marginal resection, which has minimal morbidity (5-7). It has been suggested to avoid surgery in asymptomatic cases even if the mass size is large (11). As in our study, it was reported that there were significant decreases in preoperative VAS scores after surgery (14,17). In addition, preoperative symptoms and shoulder functions recovered completely after surgery. We also recommend conservative treatment in asymptomatic cases due to high rates of postoperative complications.

Although long-term complications are rare, as in our study, temporary complications such as postoperative seroma or hematoma (4,9,10) and chronic pain (7,9,12) have been reported, especially when the resected tumor mass is large. In the literature, studies report complication rates as high as 43% (10,12,17) and as low as 10% (5,7,9,19). In our study, seroma, hematoma, and pain were observed frequently in 35% of the patients. While some researchers have stated that complications such as postoperative seroma or hematoma are more common in cases with large resected tumor masses (7,10,17,19), some have reported no differences (4). No significant complications were reported after the marginal excision of masses with a mean size of 9.6 cm (18). It is known that postoperative complications are more common in elderly patients and those with comorbidities (11). Although we did not find a significant relationship between age and complications, we observed a significant positive relationship between complications and the size of the mass, consistent with previous studies. It has also been reported that more hematoma develops in cases with large masses and shorter durations to drain removal (10), and that operative times are longer and complication rates are higher in patients with BMI of >25 (4). We think that the high complication rate seen in our study is related to early shoulder movements. In our study, no significant relationship was found among drain removal time, complications, and lesion size. This may have been due to the relatively short durations until drain removal and hospitalization time among our patients. While we statistically observed larger masses in people with higher BMI values, we did not find a significant relationship between BMI and complications. This may be because it is not easy to identify the presence of seroma/hematoma in obese individuals.

More complications were reported after synchronous surgeries performed in bilateral cases (1,12), and it was recommended that this approach only be applied in cases where masses are large and symptomatic (12). We encountered a significantly higher rate of complications in synchronous surgeries (6/9) compared to sequential surgery (1/3) among our bilateral cases. It has been reported that preoperative discomfort is increased due to mechanical irritation, postoperative care becomes more difficult (11), and sleep disorders are observed more often in patients who have a habit of sleeping in the supine position (14). In our study, significantly more complications were observed in 14/31 patients who slept in the supine position.

Recurrence after marginal resection is extremely rare (5,12) and was observed only in cases with incomplete resection (1,5) and positive macroscopic-microscopic margins (3,15). A 4.5% recurrence rate was found in a

previous study (19), but studies with higher recurrence rates also exist in the literature (8,17). For a patient who was referred to us with recurrence after surgery in another hospital, a successful result was obtained by performing excision for the recurrence. ED formation in the contralateral scapular region, which was followed conservatively, was observed in two other patients during follow-up.

Aspiration drainage (12,18,19), compressive bandages (7,9), and shoulder immobilization (5,10,12) are recommended to prevent postoperative seroma/hematoma formation. There is no consensus in the literature regarding the ideal time to drain removal (5,9), postoperative shoulder immobilization (5,10), or the duration of postoperative follow-up (1,4,5). Drains were removed based on clinical observation, with Hemovac drains used in cases with bleeding and Penrose drains in other cases. We followed the patients at short intervals for the first 6 months and then recommended annual follow-up appointments.

The limitations of our study are its retrospective and monocentric nature and the lack of a control group. However, a large number of cases, a long follow-up period, and a diverse patient population are the strengths of the study.

CONCLUSION

In symptomatic cases of ED, marginal resection with a muscle-sparing approach is a safe and effective surgical method providing satisfactory clinical results. The decision for resection should be made after a comprehensive evaluation of the symptoms and the lesion. Elderly obese patients who sleep in the supine position and have comorbidities should particularly be informed about possible complications. Careful surgery and hemostasis, prolongation of the time to drain removal and follow-up periods, and avoidance of synchronized surgery in bilateral cases are beneficial in preventing complications.

ETHICAL DECLARATIONS

Ethics Committee Approval: The study was carried out with the permission of University of Health Sciences Dr. Abdurrahman Yurtaslan Oncology Health Applications and Research Center Clinical Researches Ethics Committee (Date: 06.10.2021, Decision No: 10-1419).

Informed Consent: All patients signed the free and informed consent form.

Referee Evaluation Process: Externally peer-reviewed.

Conflict of Interest Statement: No conflict of interest was declared by the authors.

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Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the manuscript, and they have approved the final version.

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