CASE REPORTS

Lambl's excrescences and a review of therapeutic strategies

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ABSTRACT

Background: Lambl's excrescences (LEx) are thin, filiform structures that arise on the lines of closure of heart valves. Although rare, we have come across various case reports in the literature describing thromboembolic events associated with LEx. We report the case of a 63-year-old female who presented with TIA-like symptoms and was found to have a LEx on the aortic side of native aortic valve. We conducted a comprehensive literature review with emphasis on different therapeutic strategies utilized to provide insight for future encounters.

Case presentation: A 63-year-old female with a past medical history significant for hypertension and hyperlipidemia presented to the emergency room with symptoms of dysarthria and unsteady gait. While in the hospital, the patient returned to her baseline gait and speech without intervention. Work-up for including computed tomography of the head, magnetic resonance imaging, and carotid doppler, as well as prolonged telemetry monitoring was negative. A transthoracic echocardiogram showed a 0.81 cm linear echodensity attached to the aortic side of the aortic valve, highly suggestive of LEx that was later confirmed on transesophageal echocardiogram. Patient was placed on clopidogrel alone for antiplatelet therapy due to an aspirin allergy and she was subsequently discharged home.

Discussion: No standard management for LEx currently exists. We aim to add to the evidence of effective management and explore the literature of patients who have experienced TIA/CVA symptoms secondary to LEx.

Key Words: Lambl's excrescence, Cryptogenic stroke, Thromboembolism

1. BACKGROUND

Lambl's excrescences (LEx), first described by Bohemian physician Vilém Dušan Lambl in 1856, are thin, mobile, filiform structures that arise on the lines of heart valve closures.^[1–3] They are thin (up to 1.5 mm in width) and elongated, varying from 5 mm to 10 mm.^[4,5] Some excrescences, known as giant LEx's, have been noted to be up to 2.5 cm in length.^[6] There are two types of excrescences with different age population, lamellar and filiform, that were found by Hurle et al. Lamellar excrescences occurred more often in those younger than 30 years old, while filiform excrescences most often occur more frequently in persons older than 40 years old.^[7] These structures are thought to be secondary to intimal damage – the continual striking of the valve causes small areas of damage to the endothelium - allowing fibrin to cover the damaged area. This fibrin structure becomes partially uplifted from the valve surface. A layer of intimal cells than covers the fibrous tissue, making the excrescence.^[8,9] Histopathologically, these structures are a central core of elastic connective tissue, covered by a single

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layer of endothelial cells.^[2] The differential diagnosis of a mobile structure off of a heart valve includes but not limited to, LEx, papillary fibroelastoma, cardiac myxoma, infective endocarditis, thrombus, and dissection of the aorta.^[10]

LEx have been described on both native and prosthetic valves, but they are most commonly found on the atrial side of the mitral valve and the ventricular side of the aortic valve.^[11] In a study by Roldan et al., they performed echocardiography on 227 patients, and found 95 of who had LEx, and 21 of those patients had them on multiple valves. 69 patients found excrescences on the mitral valve, 41 on the aortic valve, 6 on the tricuspid valve, and 1 on the pulmonic valve.^[4] It has been reported that LEx can be found on 70%-80% of adult heart valves, and there is a similar prevalence of LEx in both healthy subjects and patients with or without suspected cardio-embolic disease, regardless of age or gender.^[4,12] Although rare, various case reports exist in literature describing thromboembolic events associated with LEx. These include transient ischemic attack (TIA), ischemic stroke, myocardial infarction, and embolization to peripheral arteries.^[6,13–15] While no standard management for LEx currently exists, various case reports describe the management of patients who have stroke-like symptoms using anticoagulation, antiplatelet, surgical excision, and conservative management.^[3, 8, 16, 17] In this report, we present a case of TIA likely secondary to embolism of LEx on the aortic side of the native aortic valve. We also conducted a comprehensive literature review with an emphasis on different therapeutic strategies utilized to provide insight for future encounters.

2. CASE PRESENTATION

The patient is a 63-year-old female with a past medical history significant for hypertension and hyperlipidemia who presented to the emergency room with symptoms of dysarthria and unsteady gait. Her symptoms started three days prior to presentation, associated with recurrent episodes of slurred speech and word finding difficulty. She did not have any focal, facial, or limb weakness. However, she suffered multiple episodes of mechanical fall due to unsteady gait. On presentation, vitals and laboratory studies were normal and physical exam was otherwise unremarkable, including a normal neurologic examination. Computed Tomography (CT) of the head and Magnetic Resonance Imaging (MRI) of the brain did not show any acute intracranial pathology. Doppler of the carotids did not demonstrate any cerebrovascular insufficiency in extracranial carotid artery system. Electrocardiogram (ECG) and continuous telemetry monitoring did not demonstrate any cardiac arrhythmia. Transthoracic echocardiogram showed borderline low ejection fraction and mild

aortic valve leaflet thickening with a linear echodensity attached to the aortic side of aortic valve that was highly suggestive of LEx. It was later confirmed on transesophageal echocardiogram (see Figure 1). No histopathology was obtained for this patient, as she was treated medically. Patient was subsequently placed on clopidogrel alone for antiplatelet therapy due to her prior aspirin allergy and the patient was subsequently discharged home with plan for close outpatient follow-up.



Figure 1. Transesophageal echocardiogram demonstrating 0.81 cm echodense structure on the aortic side of native aortic valve suggestive of LEx

3. DISCUSSION

No standard of management for LEx currently exists. A comprehensive literature review with an emphasis on the different therapeutic strategies was done, summarized below (see Table 1). Exclusion criteria included any studies where a LEx was an incidental finding and when patients were already on anticoagulation or antiplatelet therapy for other reasons. As noted previously, LEx are more commonly found on the atrial side of the mitral valve and the ventricular side of the aortic valve. However, most symptomatic patients were found to have LEx on the aortic valve,^[3,6,11,13–16,18–34] as our patient had.

Previous case reports have suggested a variety of treatment modalities, including anticoagulation, antiplatelet therapy, surgical excision, and conservative management.^[15,24,27,34-37] Medical management within itself varied greatly, ranging from aspirin alone, dual antiplatelet therapy, novel oral anticoagulants, to full anticoagulation therapy with warfarin.^[18,24,27,34,36] Roldan et al. performed a study that suggests LEx may not be a source of cardioembolic strokes, and it may be reasonable to not treat them at all.^[4] While there is no consensus on a preferred treatment modality, our literature review revealed that in five of the six studies where the patient presented with a TIA, the patients were treated with a combination of antiplatelet therapy with different agents, and only one patient was treated with surgical excision.^[13, 16, 34–36, 39] In a patient whose medical compliance may be an issue, surgical excision may be a better option for a

patient with TIA/CVA symptoms secondary to LEx given the bleeding risk on antiplatelet/anticoagulation therapy. Those patients also experienced positive outcomes, similar to those managed conservatively.^[17, 19, 21, 23]

Table 1. Case	reports invol	lving symptoma	atic Lambl's	excrescences

Reference	Patient Demographics	Location of LE	Presentation	Management	Follow-up outcome
Kalavakunta ^[3]	59 y/o M	Aortic valve	CVA	Warfarin	No further neurological deficits that hospital course
Aggarwal ^[6]	66 y/o F	Aortic valve	CVA	Anticoagulation; Surgical excision	Not reported
Voros ^[8]	72 y/o M	Pulmonic valve	CVA	Conservative management	Not reported
Kamran ^[11]	74 y/o F	Aortic valve	CVA	Anticoagulation	Not reported
Siles ^[13]	42 y/o M	Aortic valve	TIA	ASA 200 mg	No neurological event at 1 year follow-up
Mito ^[14]	50 y/o F	Aortic valve	MI	Not reported	Not reported
Fitzgerald [15]	70 y/o M	Unknown	Sudden onset of pain in the left leg – embolus to popliteal artery	Echocardiogram; no intervention	Periodic echocardiograms for 18 months – no other events noted
Chu ^[16]	51 y/o F	Aortic valve	TIA	ASA 81 mg, clopidogrel 75 mg	No neuro events at 6 months follow-up
Al-ansari ^[17]	33 y/o M	Mitral valve	CVA	Surgical excision	Not reported
Davogustto [18]	68 y/o F	Aortic valve	CVA	ASA 81 mg	No recurrence of symptoms at 2 years follow-up
Aziz ^[19]	61 y/o F	Aortic valve	AMS	Surgical excision	10 days after discharge – no neuro events noted
Quinson [20]	64 y/o F	Aortic valve	Angina	Surgical excision	No neuro events at 3 months follow-up
Berent [21]	44 y/o F	Aortic valve	Weakness and fatigue	Surgical excision	Not reported
Wolf ^[22]	47 y/o F	Aortic valve	CVA	ASA 100 mg	No neuro events at 6-month follow-up
Liu ^[23]	53 y/o M	Aortic valve	CVA	ASA 100 mg	Symptom-free at 9 months
Liu	30 y/o M	Aortic valve	CVA	Surgical excision	Symptom-free at 7-month follow-up
Wu ^[24]	66 y/o F	Aortic valve	CVA	Warfarin	No neuro event at 6-month follow-up
Yacoub ^[25]	59 y/o M	Aortic valve	CVA	ASA and Lipitor	Not reported
Raju ^[26]	44 y/o M	Aortic valve	CVA	Surgical excision; discharged on antiplatelet therapy	Not reported
Meireles [27]	56 y/o F	Aortic valve	CVA	Rivaroxaban 20 mg daily	Not reported; neurologic improvement noted
Rhee ^[28]	21 y/o M	Aortic valve	CVA	ASA 100 mg daily	No neuro event after 3 years follow-up
Pizzuti ^[29]	59 y/o M	Aortic valve	MI	Surgical excision	Not reported
Dangas [30]	75 y/o F	Aortic valve	Unstable Angina	Surgical excision	Not reported
Vlacancich [31]	39 y/o F	Aortic valve	CVA	Dual antiplatelet therapy	Not reported
Kariyanna [32]	62 y/o F	Aortic valve	CVA	ASA and high-dose statin	Not reported
Manolakis ^[33]	47 y/o F	Aortic valve	CVA	Warfarin (not adherent) Surgical excision	Experienced another stroke, secondary to cocaine use
- [34]	54 y/o M	Aortic valve	TIA	Clopidogrel and ASA	No neuro event after 2 years follow-up
JO [24]	78 y/o M	Aortic valve	CVA	Dabigatran	Not reported
Villella ^[35]	63 y/o M	Mitral valve	TIA	Antiplatelet therapy	Still having symptoms 2 months later, but less frequent
Ker [36]	32 y/o M	Mitral valve	TIA	ASA 100 mg, clopidogrel 75 mg	No neuro event after 3 years follow-up
Fumero ^[37]	30 y/o M	Mitral valve	CVA	Surgical excision	No neuro event after 1 year follow-up
Nighoghossian [38]	31 y/o M	Mitral valves	Recurrent CVA	Acenocoumarol; Surgical excision	Not reported
Bruinsma ^[39]	34 y/o M	Mitral valve	TIA	Surgical excision	Not reported

Chu et al. suggested LEx should be included in the differential diagnosis of cryptogenic stroke, and if found on TEE, should be treated with dual antiplatelet therapy.^[16] Ker et al. and Rhee et al. are two studies with the longest follow-up of three years. Both studies had young male patients; Ker's patient was treated with dual antiplatelet therapy, while Rhee's patient was treated with aspirin alone.^[28,36] The CHANCE trial suggests dual antiplatelet therapy with clopidogrel and aspirin is superior to aspirin alone for reducing the risk of stroke in patients who had experienced a TIA or minor stroke without increasing the risk of hemorrhage.^[40]

Given the clinical significance, we believe that more studies regarding the management of LEx should be done.

4. CONCLUSION

As Chu et al. suggested, LEx should be considered in the differential diagnosis of cryptogenic stroke. Although rare and under represented, it is imperative to have a consensual standard management plan for patients suffering form cardioembolic disease secondary to LEx. Using the case of our patient as an example, we aim to add to the evidence of effective management of patients who have experienced TIA/CVA symptoms secondary to LEx by successful treatment with antiplatelet therapy.

CONSENT

Consent for publication was obtained from the patient.

CONFLICTS OF INTEREST DISCLOSURE

The authors declare no conflicts of interest.

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