Fungal prosthetic mitral valve endocarditis caused by *Scopulariopsis* species: Case report and review of the literature

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S*copulariopsis* species is a keratinolytic mold with a propensity to cause onychomycosis and rarely to cause deep tissue infection. *Scopulariopsis brevicaulis* is the most common *Scopulariopsis* species seen clinically. We review 3 cases of *Scopulariopsis* species-induced endocarditis in the literature and present a fourth case of *Scopulariopsis* species-induced prosthetic valve endocarditis.

Clinical Summary

A 67-year-old woman with a past medical history significant for rheumatic fever, gout, pulmonary hypertension, atrial fibrillation, congestive heart failure, and 3 prior valve operations presented with fever and right leg pain. One year before presentation she underwent replacement of her aortic and mitral valves with a 19-mm Carpentier-Edwards Magna pericardial valve and a 25-mm Carpentier-Edwards pericardial valve (Baxter Healthcare Corp, Edwards Lifesciences, Irvine, Calif), respectively. Her social history was significant for pet finches. Her temperature was 101°F, she had a systolic murmur at the left sternal border, and her right foot was cold with diminished pulses. She had no evidence of onychomycosis or peripheral emboli. Abdominal computed tomography demonstrated a nonocclusive clot in the right ileac artery. Hematologic parameters and chemistries were normal. Blood cultures, including fungal isolator blood cultures incubated over 4 weeks, were negative. After the patient's left leg became cold and pulseless, she urgently underwent thromboendarterectomy of an acutely occluded femoral artery. Pathology from the embolectomized specimen demonstrated septated hyphal elements. Subsequently on day 5, fungal cultures grew a mold morphologically identified as a *Scopulariopsis* species (Figure 1). She underwent a transesophageal echocardiogram revealing a larger than 12-mm vegetation on the mitral valve with moderate mitral regurgitation. Confirmatory identification and susceptibility testing

Figure 1. *Scopulariopsis* species isolated from the patient's thrombectomy. (Original magnification 1000×; lactophenol cotton blue stain.)

performed at Focus Diagnostics (Cyprus, Calif) identified the organism as *S brevicaulis* and demonstrated mean inhibitory concentrations of 4 µg/mL to amphotericin B, 1 µg/mL to terbinafine, 8 µg/mL to voriconazole, and greater than 8 µg/mL to itraconazole. The patient underwent excision of the senescent infected mitral valve pericardial prosthesis, extensive debridement of the associated fungal vegetation, and a mitral valve replacement with a Carpentier-Edwards pericardial valve. She was treated preoperatively and postoperatively with amphotericin B and voriconazole. The amphotericin was continued for 3 weeks postoperatively, and the voriconazole was arbitrarily continued for 11 weeks until the patient could not tolerate it because of debilitating nausea. She had a complicated respiratory course postoperatively and was discharged to a rehabilitation facility. She was subsequently readmitted with dyspnea and died 5 months after initial presentation without signs of recurrent infection. Culture of her pet finch's feathers grew *Aspergillus* species and not *Scopulariopsis* species, despite the mold's propensity to grow in bird feathers.

Discussion

Fungi are responsible for less than 10% of cases of infective endocarditis. Although the majority of these are caused by *Candida* species, infections with opportunistic mold are seen in patients with previous valve operations.¹

In the last 10 years, prosthetic valvular endocarditis caused by *Scopulariopsis* species has been reported in 3 cases in the literature (Table 1).²⁻⁴ Major emboli, a common presentation of fungal

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TABLE 1. Four cases of prosthetic valve endocarditis caused by *Scopulariopsis* species

Case	Presentation- echocardiogram	Timing of presentation	Diagnostic study	Treatment	Outcome
Gentry and coworkers, 1995 ²	Abdominal pain, hematuria caused by large renal infarct; pedunculated vegetation on echocardiogram	Twelve months after valvuloplasty with Duran prosthetic ring	Valve culture grew <i>S brevicaulis</i>	Surgical debridement and new St Jude's mitral prosthesis; 66 days of amphotericin B and then 90 days of itraconazole	No recurrence at follow-up
Migrino and coworkers, 1995 ³	Chest pain; echocardiogram demonstrating 3 cm of vegetation on aortic prosthesis; then right leg claudication with embolism to femoral artery	Presented 11 months after Carpentier-Edward aortic valve replacement for AS	All blood cultures, including fungal isolator blood cultures, were negative; cultures of the embolus and prosthesis grew <i>S brevicaulis</i>	Treated with amphotericin B and ketoconazole for synergy; highly resistant in vitro to all agents	Progressive infection leading to death despite 2 valve operations and antifungal therapy
Chen-Scarabelli and Scarabelli 2003 ⁴	Fever, purple fingers; echocardiogram with vegetations on ventricular side of MV and linear atrial densities	Five months after her third mitral valve replacement	All blood cultures were negative; valve culture grew <i>Scopulariopsis</i> species	Bioprosthetic valve replacement of mechanical valve, treatment with caspofungin	Alive at hospital discharge
Current case	Cold leg, femoral arterial septic emboli; echocardiogram with 12 mm of mitral vegetation	One year (see above for details)	Blood cultures and fungal isolator blood cultures were negative; culture of embolus grew <i>S brevicaulis</i>	Treated preoperatively with amphotericin B and voriconazole; bioprosthetic valve replacement; treated postoperatively with 4 weeks of amphotericin and 11 weeks of voriconazole	Alive at hospital discharge; ultimately expired from pulmonary complications without recurrence

AS, Aortic stenosis; MV, mitral valve.

endocarditis,¹ occurred in 3 of the 4 reported cases, including our case. Blood cultures did not grow in any of the cases, and the diagnoses were based on fungal cultures of embolectomy tissue and prosthetic heart valves. In the cases in which susceptibility patterns were reported, the organism demonstrated significant in vitro resistance to multiple antifungal agents, corroborating with other reports noting high-level resistance in clinical isolates of *S brevicaulis*.⁵ Fungal valvular vegetations generally require surgical intervention for treatment because a long-term cure for fungal prosthetic endocarditis is unlikely. Subsequent antimicrobial therapy is typically continued for long durations. Although clinical correlation with in vitro susceptibility data is not fully established for *Scopulariopsis* species, the high level of in vitro resistance demonstrated for *Scopulariopsis* species to multiple antifungal agents,⁵ taken together with several reports of emergency and

progressive infection on broad antifungal therapy, suggests that successful treatment without surgical debridement might be even more unlikely for *Scopulariopsis* species than for other more treatment-responsive fungi.²⁻⁴ The appropriate duration of antifungal therapy after surgical intervention is unclear, although it is reasonable to continue antifungal agents for several weeks to months through endothelialization of a prosthetic valve. When *Scopulariopsis* species is identified in a clinical specimen, before susceptibility results are known, initial empiric use of intravenous amphotericin B alone or in combination with voriconazole or itraconazole is reasonable. Additional experience is needed to optimize antifungal selection in this setting, particularly as relates to combination antifungal therapy and the role of newer azoles, such as posaconazole; the echinocandins, such as caspofungin; or the allylamine terbinafine in synergy. Given the intrinsic resistance

demonstrated by *Scopulariopsis* species in vitro, early surgical debridement is the mainstay of therapy for *Scopulariopsis* species-induced endocarditis and should be strongly considered when *Scopulariopsis* species infection is diagnosed.

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Erosion of a retroesophageal subclavian artery by an esophageal prosthesis

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Supplemental material is available online

Arterioesophageal fistula is a rare cause of upper digestive hemorrhage. It usually presents with a fatal outcome. It may happen in the course of esophageal or thoracic disease, or in the setting of foreign body ingestion, and involves the thoracic aorta or the subclavian artery.

Arterioesophageal fistula can also occur in the presence of a retroesophageal subclavian artery (RESCA), also known as *arteria lusoria*. It is usually asymptomatic but may cause dysphagia (*dysphagia lusoria*) and may demonstrate symptomatic atherosclerotic and aneurysmal degeneration. However, spontaneous fistulization in the esophagus is not described. It has mostly been associated with prolonged nasogastric tube presence. The present report describes the first case of arterioesophageal fistula with a RESCA caused by an esophageal prosthesis and discusses treatment based on a review of the literature.

Clinical Summary

Fourteen months before the actual admission, a 9-year-old girl with Down syndrome was admitted for caustic ingestion. She presented with severe ulceration from the middle esophagus down to the stomach. The acute episode was treated conservatively. Nonetheless, dysphagia rapidly developed in the patient. Two months after the accident, an esophagoscopy disclosed active stenotic esophagitis of the last third of the esophagus. Iterative pneumatic dilatations were performed, which were discontinued because of esophageal fissuration and mediastinitis. A 14-mm Polyflex stent was inserted that rapidly migrated into the stomach; it was extracted and replaced by a 16-mm stent.

In June 2005, the patient was admitted for hematemesis and melena. Emergency esophagoscopy showed blood oozing around the prosthesis. The endoscopic procedure was interrupted, and the child was brought to the catheterization laboratory where the prosthesis could be mobilized under angiographic control. When the prosthesis was pushed toward the stomach, a major hemorrhage occurred and a big aortic branch was identified as the source of bleeding. First it was regarded as a major bronchial artery. The prosthesis was pulled back to enable partial control of the bleeding, and coil embolization was performed in the so-called bronchial artery. This maneuver did not control the hemorrhage, and a RESCA was identified as the source of bleeding (Figure 1). RESCA embolization was attempted unsuccessfully, despite satisfactory angiographic images (Figure 2), and the patient presented sustained bleeding with hemorrhagic shock and cardiac arrest. After successful reanimation the patient underwent emergency surgery consisting of ostial RESCA ligation while the endoesophageal prosthesis was left in place. An 8-mm polytetrafluoroethylene (Gore-Tex, Flagstaff, Ariz) prosthetic bypass was constructed between the lateral aspect of the ascending aorta and the axillary artery. The patient had a prolonged intensive care unit stay (23 days) but fully recovered and was discharged without after-effects. Although the first esophagoscopy control showed embolized coils visible through the ulcerated esophageal mucosa (Figure E1), the lesions finally healed.

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