Case Reports

Intracardiac Abscess with Cutaneous Fistula

Secondary to Ventricular Septal Defect Repair Simulating Sternal Wound Infection

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Cutaneous fistula as a clinical presentation of intracardiac abscess of the right side is such an unusual occurrence that it has not until now been reported in the English-language medical literature. We present a rare case of right-sided infective endocarditis caused by Achromobacter xylosoxidans *in which recurrent infection presented as sternal wound discharge. The infection was found to have an intracardiac origin and was successfully managed by radical débridement on cardiopulmonary bypass.* ***(Tex Heart Inst J 2014;41(3):324-6)***

Superficial and deep wound infections are well-known complications after open-heart surgery. Right-sided endocarditis in non-drug users is uncommon, comprising 5% to 10% of infective endocarditis (IE) cases.[1] Most of these (55%) occur in patients with congenital heart disease[2]; and *Streptococcus viridans* causes 55% to 65% of these infections, whereas *Staphylococcus aureus* predominates in drug users. Intracardiac abscess as a presentation of IE is frequently related to a periannular extension and is seen in 14.4% of IE cases.[3] Cutaneous fistula as a consequence of infection after intracardiac repair is a rare entity. We report an unusual case of intracardiac abscess due to *Achromobacter xylosoxidans* infection many years after repair of a perimembranous ventricular septal defect (VSD). The only clinical manifestations were a cutaneous fistula and sternal wound discharge.

Case Report

A 50-year-old woman had undergone VSD repair at age 9 years and again at age 16, followed by surgery for endocarditis at age 45. At that time, she had vegetations on the pulmonary valve and in the right ventricular outflow tract (RVOT) and was treated with débridement of all vegetations and repair of the tricuspid valve, followed by 6 weeks of intravenous antibiotic administration. We did not consider removal of the VSD pledgets at that juncture and hoped that the treatment was curative. Cultures were positive for *Achromobacter* (*Alcaligenes*) *xylosoxidans*. Recovery was uneventful. Subsequently, the patient underwent splenectomy for pancytopenia and splenomegaly. Pathologic evaluation of the spleen showed atypical lymphoid infiltrates.

Four years later, she started having pain and purulent discharge from the mid-sternal wound. Computed tomography (CT) revealed a well-healed sternal bone without evidence of osteomyelitis but showed a soft-tissue mass adjacent to the sternal wires. She underwent local wound débridement and sternal wire removal twice within a few months and was treated with antibiotics. Despite antibacterial maintenance therapy, the fistula recurred. Repeat CT of the chest and echocardiograms did not reveal any deep disease process; nevertheless, the patient was returned to the operating room a 3rd time.

Because exploration this time disclosed that the fistulous tract originated from the heart and necessitated sternotomy, further exploration was performed with the patient on cardiopulmonary bypass. When we followed the tract into the right ventricle, we found the infection to be an abscess around the pledgets of the original VSD repair. Complete débridement of all infected material and inflammatory tissue surrounding the fistula was performed, with preservation of the septum and the tricuspid valve (Figs. 1 and 2). Removal of the pledgets did not reopen the VSD—indeed there seemed to be a solid, fibrous tissue membrane (capsule) toward the left side. Cultures again grew *A. xylosoxidans*. The patient was treated with intravenous piperacillin/tazo-

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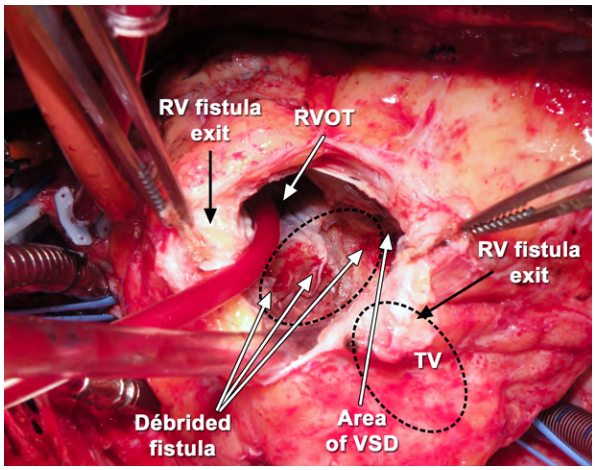

Fig. 1 *Intraoperative photograph shows the right ventriculotomy, performed to trace the fistula tract.*

RV = right ventricular; RVOT = right ventricular outflow tract; TV = outline of tricuspid valve; VSD = ventricular septal defect

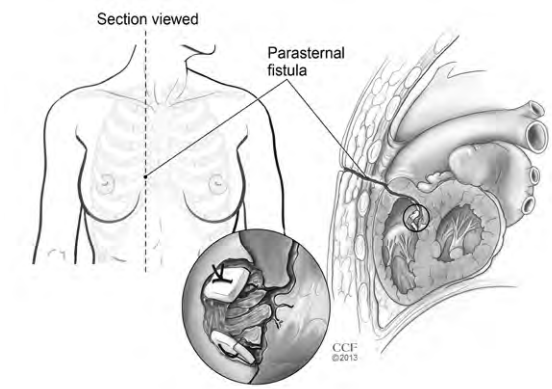

Fig. 2 *Illustration depicts the course of the fistula tract and shows the pledgets, which were encased in an inflammatory abscess.*

bactam for 8 weeks and was prescribed trimethoprim/sulfamethoxazole 2 times daily indefinitely, for suppression of bacterial activity.

Upon primary healing of the sternal wound, our patient's postoperative course was uneventful. Intraoperative and postoperative echocardiograms revealed no residual VSD or tricuspid valve regurgitation. As of December 2013, more than 3 years after her discharge from the hospital, she had a well-healed sternal wound without any evidence of recurrence and was leading an active, healthy life.

Discussion

This case illustrates that an infection originating in prosthetic material can persist, can be dormant for extended periods of time, and can in essence be incurable with antibiotics alone.[4] During this patient's surgical excision of vegetations in the RVOT, the true origin of the infection was not identified; after another 5 years, the infection re-manifested itself with external discharge from the mid sternum. In the absence of our finding and treating the source of the infection, our local débridement and sternal wire removals failed. Following the infection down to the right ventricle and radically débriding all foreign material (which included the pledgeted sutures from the VSD) resolved the problem. A thick fibrous capsule prevented recurrence of the VSD.

Achromobacter xylosoxidans is an opportunistic pathogen and is probably part of the endogenous flora of the ear and gastrointestinal tract. It is an aerobic, motile, gram-negative rod bacterium first described in 1971 by Yabuuchi and Oyama,[5] who discovered it in patients with chronic otitis media. Nosocomial outbreaks have been attributed to disinfectant solutions, saline solution, and diagnostic tracers contaminated with this organism, which typically inhabits aqueous environments.[6] Few cases of IE due to *A. xylosoxidans* have been published. This opportunistic pathogen is difficult to eradicate[7] because of its unusual antibiotic-susceptibility profile, which includes high levels of resistance to cephalosporin, aminoglycoside, and quinolone.[8] Drug resistance in *A. xylosoxidans* appears to be mediated secondary to plasmids produced by the bacterium.[9] Minocycline, imipenem, meropenem, piperacillin, and piperacillin/tazobactam display the highest activity against the organism.[10] *Achromobacter xylosoxidans* is an emerging pathogen in catheter-related infections among the dialysis-dependent population[11] and in immunocompromised hosts.[12]

In summary, we describe a very rare presentation (with a cutaneous fistula as the only clinical manifestation) of an intracardiac abscess caused by *A. xylosoxidans*. That this occurred many years after repair of a perimembranous VSD serves as a reminder that only radical surgical débridement can cure some infections, especially when they are associated with an indolent pathogen such as this one.

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