Case Report

Infected abdominal aorta aneurysm secondary to streptococcal toxic shock syndrome due to Streptococcus pyogenes: a case report from Japan

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Background: Infected aortic aneurysm secondary to streptococcal toxic shock syndrome caused by *Streptococcus pyogenes* is uncommon and associated with high mortality.

Case presentation: A 75-year-old man with metastatic lung cancer and an abdominal aortic aneurysm presented with high fever for 3 days. He was diagnosed with septic shock and was admitted to our hospital. The blood culture was positive for *S. pyogenes*, and streptococcal toxic shock syndrome was diagnosed. During treatment, enhanced computed tomography revealed an increase in the size of the abdominal aortic aneurysm, leading to the diagnosis of an infected aortic aneurysm. Replacement of the aneurysm with a synthetic graft was carried out successfully. The patient gradually recovered after the surgery.

Conclusion: We successfully managed an infected aortic aneurysm secondary to streptococcal toxic shock syndrome. Infected aortic aneurysms should be considered in patients with a medical history of aortic aneurysms and presenting with streptococcal toxic shock syndrome.

Key words: Cancer, group A streptococcus, infected abdominal aortic aneurysm, streptococcal toxic shock syndrome, Streptococcus pyogenes

INTRODUCTION

STREPTOCOCCAL TOXIC SHOCK syndrome (STSS), caused by *Streptococcus pyogenes* (*S. pyogenes*), is associated with critical complications and mortality.[1] Infected aortic aneurysms (IAAs), although uncommon, are also associated with severe complications and mortality.[2] Although *S. pyogenes* is not a major causative pathogen of IAAs,[3] 13 cases of *S. pyogenes*-related IAAs have been reported.[4–16] Herein, we report the case of a patient with an asymptomatic infected abdominal aortic aneurysm who successfully underwent aortic replacement with a bifurcated vascular prosthesis following *S. pyogenes*-induced STSS.

CASE

A 75-YEAR-OLD MAN was transferred to our hospital by the emergency medical service with a complaint of high fever (>39.0°C) for 3 days. He had a history of metastatic lung cancer following a hypopharyngeal cancer surgery and was treated with nivolumab. He also had a 25-mm abdominal aortic aneurysm involving the iliac bifurcation. Vital signs showed altered mental status (Glasgow Coma Scale score, 14 [E4V4M6]) and shock (heart rate, 117 b.p.m.; blood pressure, 66/47 mmHg). Physical examination revealed erythema on the left shoulder. Hypoxia and hypotension necessitated mechanical ventilation, continuous norepinephrine administration, and vasopressin infusion. Laboratory investigations revealed thrombocytopenia, elevated creatinine, coagulopathy, liver dysfunction, and abnormal serum lactate levels (Table 1). A computed tomography (CT) scan of the trunk did not show any radiological signs

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Table 1. Laboratory findings on day 1 of admission of a 75-year-old man with infected abdominal aorta aneurysm secondary to streptococcal toxic shock syndrome

Arterial blood gas analysis (FiO_2, 0.4)			Biochemistry		
pH	7.480		Total protein	5.3	g/dL
pCO_2	21.0	mmHg	Albumin	2.6	g/dL
pO_2	120.0	mmHg	Na	126.4	mEq/L
HCO_3^-	22.4	mmol/L	K	3.9	mEq/L
Anion gap	15.6	mmol/L	Cl	95.0	mEq/L
Lactate	5.0	mmol/L	UN	33.2	mg/dL
Complete blood count			Cr	2.04	mg/dL
WBC	14.6	$\times 10^3/\mu L$	UA	7.2	mg/dL
Hb	12.3	g/dL	AST	640	U/L
Platelet	108	$\times 10^3/\mu L$	ALT	130	U/L
Coagulation			LD	1,889	U/L
PT	12.3	sec	ALP	202	U/L
APTT	38.5	sec	γ-GTP	32	U/L
Fibrinogen	578	mg/dL	AMY	212	U/L
FDP	109.6	μg/mL	CK	49,565	U/L
D-dimer	71.2	μg/mL	CRP	22.31	mg/dL
Endocrine			Procalcitonin	88.08	ng/mL
Glucose	142	mg/dL			

γ-GTP, γ-glutamyl transferase; ALP, alkaline phosphatase; ALT, alanine aminotransferase; AMY, amylase; APTT, activated partial thromboplastin time; AST, aspartate aminotransferase; Cl, chloride; CK, creatine kinase; Cr, creatinine; CRP, C-reactive protein; FDP, fibrin degradation products; Hb, hemoglobin; K, potassium; LD, lactate dehydrogenase; Na, sodium; UA, uric acid; UN, urea nitrogen; WBC, white blood cells.

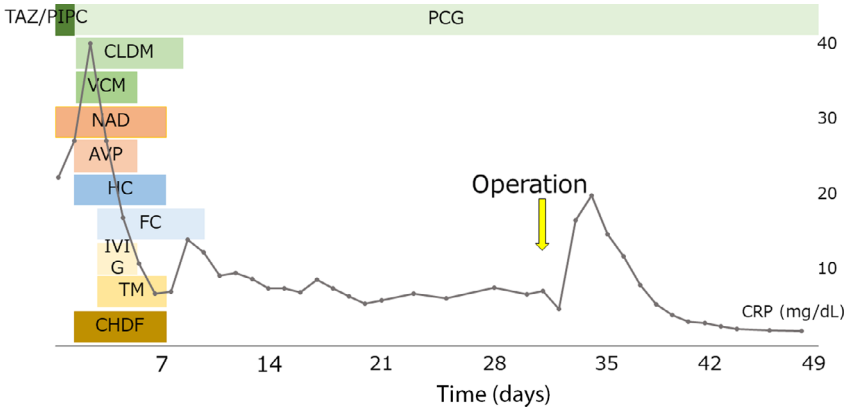

Fig. 1. Clinical course of infected abdominal aorta aneurysm secondary to streptococcal toxic shock syndrome in a 75-year-old man, with C-reactive protein levels (CRP) and treatment. AVP, vasopressin; CHDF, continuous hemodiafiltration; CLDM, clindamycin; FC, fludrocortisone; HC, hydrocortisone; IVIG, i.v. immunoglobulin; NAD, noradrenaline; PCG, penicillin G; PIPC, piperacillin; TAZ, tazobactam; TM, thrombomodulin; VCM, vancomycin.

indicative of the cause of fever. The patient was diagnosed with septic shock and was admitted to the intensive care unit on a regime of tazobactam/piperacillin. On day 2, the blood culture was positive for group A streptococcus, prompting a diagnosis of STSS. Accordingly, the antibiotics were changed to penicillin G, clindamycin, and vancomycin. Furthermore, hydrocortisone, fludrocortisone, i.v. immunoglobulin, and thrombomodulin were also given for 6, 7, 3, and 5 days, respectively (Fig. 1). Continuous hemodiafiltration with an acrylonitrile-co-methallyl sulfonate surface-treated (AN-69ST) membrane was undertaken for 6 days. Finally, the group A streptococcus was identified as

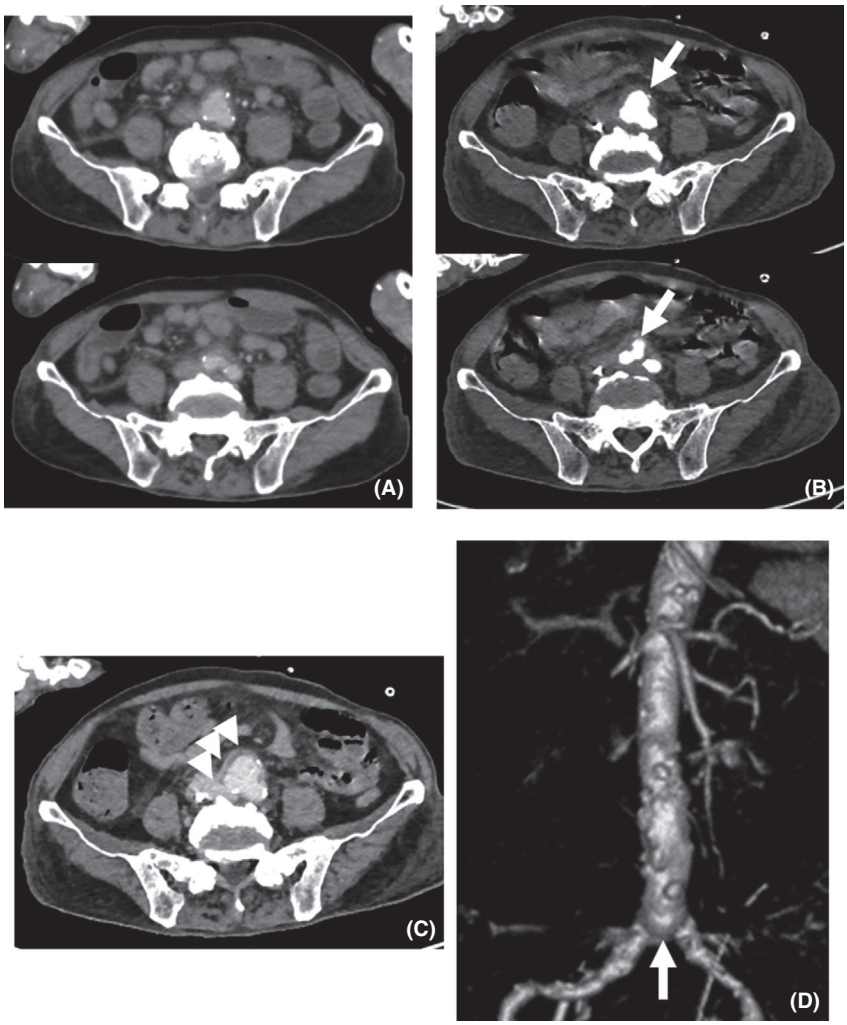

Fig. 2. Enhanced computed tomography images of an infected abdominal aorta aneurysm secondary to streptococcal toxic shock syndrome in a 75-year-old man. Images show the aortic aneurysm on day 2 (A) and day 30 (B–D). Arrows (B,D) indicate the increased size of the aneurysm compared to the size noted in the previous computed tomography examination. Arrowheads (C) indicate the enhanced aneurysm wall and saccular appearance of the aneurysm.

S. pyogenes; genetic analysis classified it as the *emm1* strain of *S. pyogenes* (T serotyping, T1; M serotyping, M1; and emm typing, *emm1*; Spegene, speA, speB, and speC). Despite treatment, the C-reactive protein levels did not decrease below 8 mg/dL (Fig. 1). An enhanced CT scan of the trunk on day 30 revealed that the aneurysm had increased in size since the previous CT scan and also revealed an enhanced aneurysm wall and a saccular appearance of the aneurysm, suggesting IAA (Fig. 2). In situ repair with a bifurcated expanded polytetrafluoroethylene vascular graft and omental wrapping were undertaken successfully on day 31. Intraoperatively, the tissue surrounding the aortic aneurysm was observed to be edematous and hemorrhagic and had strong adhesions. Culture of the sample collected from the aortic wall during surgery was negative. Furthermore, preoperative blood cultures on days 7, 14, and 31 were also negative. After the surgery, the patient recovered and the C-reactive protein level gradually reverted to normal. On resolution of ventilator-associated pneumonia, hospital-acquired pneumonia, and pneumothorax, he was weaned off the ventilator on day 189 and walked without support from day 240.

DISCUSSION

WE SUCCESSFULLY MANAGED this case of an IAA related to STSS. Both conditions have high

Table 2. Previously published cases of infected aortic aneurysm by *Streptococcus pyogenes*

Case	First author	Year of publication	Age/ sex	Site of aneurysm	Surgical treatment	Medical treatment (empiric → directed)	Outcome	*S. pyogenes* in culture		
								Blood	Aneurysm wall	Other
1	Valero	1992	65/M	Infrarenal abd	Resection with bypass graft (1HD)	ABPC/SBT, AZT → NFPC, PCG	Dead (2HD)	+	+	Throat swab +
2	Sing	1994	58/F	Infrarenal abd	Graft	N/D	Survived	−	N/D	Thrombus in aneurysm +
3	Bisognano	1997	36/M	Subclavian	Repair with graft (2HD)	N/D	Dead (8HD)	+	+	N/D
4	Barth	2000	1.5/F	Ascending	Resection with graft (8HD)	CXM, GM → PCG	Survived	+	N/D	Pericardial effusion +
5	Chen	2008	81/M	Abd	None	N/D	Dead (1HD)	+	N/D	N/D
6	Leiva	2009	63/F	Thorac-abd	Resection with bypass graft	VCM, IPM → PCG	Survived	N/D	N/D	N/D
7	Vallejo	2011	63/F	Thorac-abd	Resection with graft	VCM, IPM → PCG, DBECPCG	Survived	N/D	N/D	N/D
8	Hoffman	2012	2/M	Descending	Repair with graft (3HD)	CTRX, ABPC/SBT → CLDM, ABPC	Survived	+	+	N/D
9	Gardiner	2013	60/M	Infrarenal abd	EVAR and bypass	VCM, CTRX, MNZ → DBECPCG, PIPC/TAZ, AMPC/CVA	Survived	+	N/D	Periaortic mass +
10	Biswas	2013	58/M	CIA bif	Bypass with graft	AMPC/CVA	Survived	N/D	+	N/D
11	Tamenishi	2013	63/M	Arch and abd	Bypass (15HD) and resection (22HD)	AMPC, CLDM	Survived	+	N/D	N/D
12	Cherbanyk	2017	69/F	Infrarenal abd	Resection with graft	PIPC/TAZ → CTRX, CLDM	Survive	+	−	N/D
13	Someili	2019	70/M	Arch	EVAR	CEZ → PCG	Survive	+	N/D	N/D
14	Taniguchi	2020	75/M	Abd	Graft	PIPC/TAZ, VCM, CLDM → PCG	Survive	+	−	Sputum +

−, negative; +, positive; Abd, abdominal aorta; ABPC, ampicillin; AMPC, amoxicillin; Arch, arch aorta; Ascending, ascending aorta; AZT, aztreonam; CEZ, cefazolin; CIA bif, common iliac artery bifurcation; CLDM, clindamycin; CTRX, ceftriaxone; CVA, clavulanate; CXM, cefuroxime; DBECPCG, benzylpenicillin; Descending, descending thoracic aorta; EVAR, endovascular aneurysm repair; F, female; GM, gentamicin; HD, hospital day; IPM, imipenem; M, male; MNZ, metronidazole; N/D, no data; NFPC, nafcillin; PCG, penicillin G; PIPC, piperacillin; SBT, sulbactam; Subclavian, subclavian artery; TAZ, tazobactam; Thorac-abd, thoracoabdominal aorta; VCM, vancomycin.

mortality rates. In accordance with the existing definition,[17] STSS was diagnosed on the basis of the presence of hypotension, elevated creatinine levels, thrombocytopenia, and skin rashes. The major causes of STSS are viral infections, pharyngitis, or soft tissue infections;[18] however, these were not evident in our case.

Regarding the diagnosis of IAAs, aneurysms with sterile cultures are diagnosed as IAAs only if they appear typically eccentric and perforated or penetrated during the surgery and if patients show signs of infection requiring antibiotic therapy before surgery.[19] Among the previously reported 13 cases of IAA due to *S. pyogenes*, six cases reported specimen culture results, and five of these were positive (Table 2). Although we had no pathological evidence that suggested that the expanding aortic aneurysm was due to an *S. pyogenes* infection, the CT and intraoperative findings were compatible with those of an infected abdominal aorta aneurysm secondary to an *S. pyogenes* infection.

There are no standard strategies for treating IAAs. The optimal management of infected aneurysms, including the duration of the preoperative antibiotic therapy, remains unclear.[19] To reduce the risk of fatality, we carried out the surgery immediately after the diagnosis and in situ reconstruction with a prosthetic graft and omental wrapping, as well as lifelong antibiotic therapy.[3]

The *emm1* strain of *S. pyogenes* is the most predominant genotype in STSS.[20] Among the previously reported 13 cases, genetic information was presented in two cases; in one case, M and T serotyping revealed M3 and T3, respectively,[4] whereas in the other case, emm typing revealed the *emm44* strain,[11] indicating that different strains of *S. pyogenes* can cause IAAs. Therefore, factors contributing to its invasiveness and virulence remain unclear and warrant further investigation.

CONCLUSION

WE SUCCESSFULLY MANAGED an unusual case of IAA secondary to STSS caused by the *emm1* strain of *S. pyogenes*. Infected aortic aneurysms should be considered as differential diagnoses in cases of STSS in patients with a medical history of aortic aneurysm.

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DISCLOSURE

Approval of the research protocol: N/A.
Informed consent: Informed consent was obtained from the patient.
Registry and the registration no. of the study/trial: N/A.
Animal studies: N/A.
Conflict of interest: None.

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