Case Report

Mycotic Aneurysm of the Abdominal Aorta due to Group B Streptococcus (GBS) Infection: A Case Report and Review of the Literature

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We present a case report of an extremely rare infected abdominal aortic aneurysm (AAA) caused by group B streptococcus (GBS). A 66 years old, afebrile male patient with hypertension and history of chronic smoking presented with mild abdominal pain and discomfort. Physical examination revealed a pulsatile mass of 5 cm in diameter located at the periumbilical region. A computed tomography (CT) scan of the whole abdomen exhibited an outpouching lesion with intramural hematoma located at the infrarenal abdominal aorta, sized 5.1x3.7 cm in diameter, suggesting an abdominal aortic aneurysm. A transthoracic echocardiography demonstrated normal endocardium without vegetation. Surgical abdominal aortic aneurysm repair (aneurysmorrhaphy) was successfully done and intravenous amoxicillin/clavulanic acid was given. Tissue culture obtained from the aneurysm sac and para-aortic lymph nodes revealed group B streptococcus infection. Intravenous amoxicillin/clavulanic acid was continued for 14 days, and was transitioned to oral amoxicillin 3 grams/day. Currently, the patient has been followed-up for 20 months with neither recurrent infection nor post-operative complication. This is the first case report of infected abdominal aortic aneurysm due to GBS in Thailand.

Keywords: *Group B streptococcus (GBS), Mycotic aneurysm, Infected aortic aneurysm, Abdominal aortic aneurysm*

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Group B streptococcus (GBS) is a facultative anaerobic, gram-positive coccal bacteria arranged in chains, capable of causing infections both in humans and animals. Infections in humans are mostly subjected to vulnerable groups such as pregnant women, neonates, elderly patients, and immunocompromised hosts which could lead to serious illnesses and, eventually, death. Major risk factors for GBS infection also include chronic alcoholism and diabetes. In normal healthy individuals, GBS could be found colonizing in 30% of the gastrointestinal and genitourinary tracts[1]. A large variety of clinical syndromes have been associated with GBS infection such as urinary tract infection, skin and soft-tissue infections, osteomyelitis, endocarditis, bacteremia without focus, neonatal meningitis, and rarely pneumonia[2]. An infection in the cardiovascular system due to the GBS, other than endocarditis, is quite rare. A mycotic aneurysm or infected aneurysm of the aorta due to GBS infection is rarely documented and difficult to be diagnosed. The diagnosis is based on imaging; a computed tomography (CT) scan with contrast is usually the investigation of choice. The first documented aortic aneurysm due to GBS infection was published in 1989, due to direct extension from osteomyelitis of the L2/L3 vertebrae and paravertebral abscess[3]. Few cases have been reported following the aforementioned but has never been documented before in Thailand. The author demonstrates a very rare case of mycotic aneurysm of the infrarenal abdominal aortic aneurysm from GBS infection clinically presented in an afebrile male with abdominal pain. As far as the author concerns, this is the first case report in Thailand.

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Case Report

A 66-years-old, afebrile Thai male presented with abdominal pain and distention 2 weeks prior to admission. The patient complained of a dull aching pain confined at the epigastrium and generalized abdominal discomfort. He had an underlying disease of well-controlled hypertension. The patient further revealed a significant 5-kilograms weight loss within 1 month and history of cigarette smoking 15 packs-a-year. He was previously healthy but had never been

performed annual physical check-up. Physical examination showed a normal body temperature of 37.0°C, regular heart rate 110 beat/min, blood pressure 150/90 mmHg, and respiratory rate of 24 breaths/min. He looked very ill, and the abdominal examination unveiled normoactive bowel sounds, mild generalized tenderness with no rebound tenderness. A suspected pulsatile mass-like lesion size 5x5 cm at the periumbilical area was detected. No abdominal guarding, rigidity, hepatosplenomegaly, or ascites was observed. Cardiovascular, respiratory, and neurological examinations were unremarkable. Complete blood count revealed hemoglobin 11.7 g/dL, WBC 22,100 cells/mm^3 (PMN 91.3%, L 4.9%, Mo 3.8%), platelet count 265,000/mm^3. Random capillary blood glucose concentration was 97 mg/dL. Renal function and urine examination were normal. The C-reactive protein (CRP) level was at 187.4 mg/L (normal value <5.00 mg/L) and Erythrocyte sedimentation rate (ESR) was at 116 mm/hr (normal value 0-15 mm/hr). Chest x-ray revealed only borderline cardiomegaly with no abnormal infiltration.

Sepsis, with intra-abdominal infection, concurrent with impending rupture of the infected abdominal aortic aneurysm was considered by the medical team. Thus, two hemocultures were taken and empirical antibiotic ciprofloxacin treatment was administered due to suspected patient's cefazolin allergy. A whole abdomen CT scan with contrast (Fig. 1) revealed an outpouching lesion with the intramural hematoma at the infrarenal abdominal aorta, measured about 5.1x3.7 cm in diameter suggestion of abdominal aortic aneurysm with no osteomyelitic lesion seen. Emergency abdominal aortic aneurysm repair (aneurysmorrhaphy) was scheduled. A preoperative transthoracic echocardiography revealed no vegetation and good left ventricular ejection fraction. Intraoperative findings showed a large saccular aneurysm at the aortic bifurcation size 8 cm in diameter. An enlarged aortic lymph node was also observed. Tissue culture from both the aneurysm sac and para-aortic lymph nodes were sent due to suspected infected aneurysm. There was no immediate post-operative complication. Tissue culture obtained from the aneurysm sac and para-aortic lymph nodes revealed group B streptococcus infection. However, the hemoculture result came back negative. Antibiotic amoxicillin/clavulanic acid had been given intravenously for two weeks and the patient was discharged with oral amoxicillin 3 grams/day. Four months later, the patient was admitted for the surgical fenestrated endovascular aneurysm repair (EVAR) with stent graft on the aorta where the infrarenal aneurysmorrhaphy had previously been performed. There was no post-operative complication which was also confirmed by a subsequent, 2 months later, computed tomography angiography (CTA) imaging of the whole aorta. The patient laboratory data is shown in Table 1. Currently, the patient has been followed-up for 20 months without any recurrent infection. The patient is able to regain his full daily activities.

Discussion

In 1885, Osler used the term 'mycotic' loosely to describe any types of infection caused by fungi, bacteria, or viruses. Nowadays, 'mycotic' has been used primarily to describe fungal infections. Therefore, to prevent any confusion, the term 'infected aneurysm' is used in this report[4,5]. The source of infection in infected aneurysms can generally be differentiated into either intravascular or extravascular. Primary infected aneurysms often occur from spreading, either direct invasion or lymphatic system, from adjacent areas of infection or trauma. Pre-existing abdominal aortic aneurysms (AAA) and post-traumatic false aneurysms are susceptible to be secondarily infected. Secondary

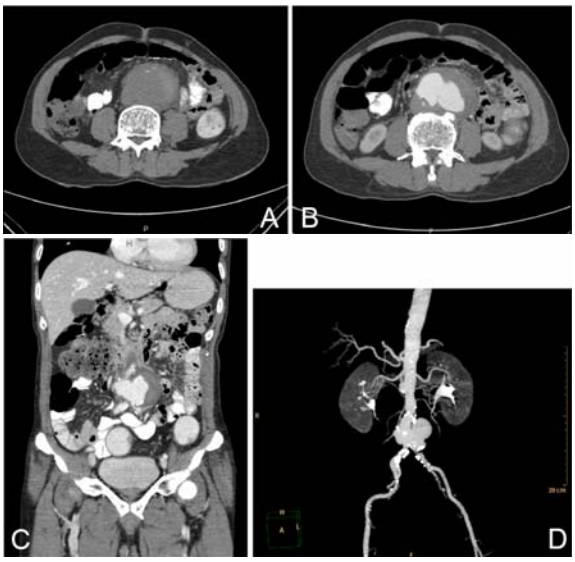

Fig. 1 Computed tomography (CT) scan of the whole abdomen with contrast shows an outpouching lesion with intramural hematoma at the infrarenal abdominal aorta, measuring about 5.1x3.7 cm in diameter suggestive of abdominal aortic aneurysm; precontrast phase (A), arterial phase (B), coronal venous phase (C), and three-dimensional reconstructed image (D).

Table 1. Laboratory results at patient presentation and during patient's follow-up

Lab (normal range)	At presentation	At discharge (2 weeks)	Follow-up (2 months)	Follow-up (20 months)
Hemoglobin (13-18 g/dL)	11.7	11.9	12.3	13.6
Hematocrit (39-52%)	35.1	35.2	36.4	40.4
White blood cell (4.4-10x 10^3)	22,100	11,120	7,350	6,800
Neutrophil (%)	91.3	72.6	62.5	62.5
Platelet (140-450x10^3)	265,000	484,000	336,000	270,000
Creatinine (mg/dL)	0.83	1.03	0.97	1.31
Alanine aminotransferase (10-20 U/L)	65	-	-	10
ESR (0-15 mm/hr)	116	65	55	30
CRP (<5 mg/L)	187.4	39.5	5.50	1.36

infected aneurysms are caused from threw septic emboli through either the vasa vasorum or intravascularly to abnormal intima areas i.e. pre-existing aneurysms or atherosclerosis. Nonetheless, organisms can sometimes colonize on the intact vessel wall through the vasa vasorum. The infected arterial wall is weakened from a local inflammatory process which consequently ends up in arteritis and aneurysm formation[6].

The incidence of an infected aneurysm is speculated to account for 0.65% to 1.3% of all aortic aneurysms[7]. Before the introduction of antibiotics, most infected aneurysms originated from bacterial endocarditis, commonly affecting the ascending or arch of the aorta. The most common pathogens were Viridans streptococci, *Enterococcus faecalis*, *Staphylococcus aureus*, *Staphylococcus epidermidis*, *Haemophilus*, and *Pneumococcus*. After the widespread use of antibiotics, the incidence of bacterial endocarditis was dramatically reduced[8]. However in non-endocarditis bacteremia, the most commonly affected site of an aneurysm is the abdominal aorta due to infected atherosclerotic plaques like in the reported patient. The most common pathogens are *Salmonella* spp. *Streptococcus*, *Bacteroides*, *Escherichia coli*, and *Staphylococcus aureus*. Other less common pathogens are also found such as *Treponema pallidum*, *Mycobacterium tuberculosis*. Fungal infections caused by *Candida, Aspergillus*, and *Histoplasma capsulatum* are quite rare but primarily found in immunocompromised hosts[6]. Concerning our case report, infection of the infrarenal abdominal aortic aneurysm is found in 95% of all AAA[9,10], though the group B streptococcus is an extremely rare pathogen causing AAA. Only a few cases have been reported so far and neither is seen in Thailand[3,11-16].

GBS or *Streptococcus agalactiae* is a facultative anaerobic, gram-positive coccal bacteria in chains. Generally, GBS does not cause serious infections in healthy hosts. However, it is best known for postpartum infections and is the most common cause of neonatal sepsis. Nowadays, it is increasingly recognized as a cause of infections in the elderly. GBS is found to be colonized in the gastrointestinal and genitourinary tract and it is estimated that 30% of the general population are asymptomatic carriers. GBS infections those are able to develop to life-threatening conditions are mostly found in neonates, elders, immunocompromised hosts, diabetes, cirrhosis, alcoholism, and cancer patients[1]. The patient in this report had none of the aforementioned risks. GBS is a re-emerging disease capable of causing a variety of clinical syndromes such as urinary tract infections, skin and soft tissue infections, osteomyelitis, neonatal meningitis, endocarditis, bacteremia without focus, and in rare cases; pneumonia and adult meningitis[2]. After a literature review on English reports of infected abdominal aortic aneurysms, only seven published reports were found. The first case reported ever was in 1989 and the latest in 2014. As mentioned earlier, neither a case has been reported before in Thailand. Information regarding a review of previous case reports of infected aneurysms is shown in Table 2[3,11-16]. Briefly, most of the reports observed that the age of patients was older than 60 years old, except for Andreasen et al. and Yamamoto et al. Infective endocarditis was not seen and the primary source of infection could not be identified in the majority of cases. Nonetheless, the overall prognosis is promising and patients showed great recovery after treatment. The most common presentation of the infected abdominal aortic aneurysm was the fever with others systemic manifestations.

Table 2. Review of the previous case reports published in English of infected abdominal aortic aneurysm to group B streptococcus

Authors	Blackett et al[3] (1989)	Andreasen et al[11] (2001)	Chandrikakumari et al[12] (2008)	Yamamoto et al[13] (2009)	Thawait et al[14] (2012)	Cozijnsen et al[15]** (2013)	Ledochowski et al[16] (2014)	Our patient (2016)
Characteristics								
Age	61	40	69	50	74	66	86	66
Sex	Male	Male	Male	Male	Male	Male	Male	Male
Complaints	Lower back pain (Lumbar)	Abdominal pain	Abdominal pain	Low back pain	Low back pain and left leg swelling	Abdominal pain and 7-kg weight loss	Abdominal pain for 10 days	Abdominal pain
Risk factors or comorbidity	No data	History of PDA correction in childhood	Diabetes mellitus, alcohol, hypertension, dyslipidemia	No data	Smoking 60-packs-a-year, hypertension, dyslipidemia	Smoking 53-packs-a-year, alcohol, hypertension, dyslipidemia	Steroid, Rituximab, cyclophosphamide for treatment Waldenstrom's disease	Smoking 15-packs-a-year, hypertension
Fever	Yes (37.5°C)	Yes	No (36.8°C)	Yes (37.8°C)	Yes	Yes (39.0°C)	Yes (38.8°C)	No (37.0°C)
Primary site of infection	Osteomyelitis at Lumbar 2-3	Unknown	Infected epidermal inclusion cyst	Unknown	Unknown	Unknown	Unknown	Unknown
Blood culture	Negative	Negative	Not done	Positive	Negative	Positive	Positive	Negative
Tissue culture*	GBS	GBS	GBS	No data	GBS	No data	Not done	GBS
Infective endocarditis	Not seen	Not seen	Not seen	No data	Not seen	Not seen	Not seen	Not seen
Antimicrobial treatment and durations	Benzylpenicillin 2 week and erythromycin 8 week	Cefuroxime 4 week	Benzylpenicillin 2 week, ceftriaxone 4 week, and moxifloxacin 18 week	Meropenem and vancomycin 3 days, cefazolin 8 days, benzylpenicillin 12 days	Gentamycin-ceftriaxone 2 week and then changed to moxifloxacin oral life long	Amoxicillin iv 12 g/day and gentamicin 3 mg/kg/day for 3 week; postoperatively: benzylpenicillin 2 week	Amoxicillin 4 week and gentamicin 2 week then ceftriaxone and clindamycin 3 week then amoxicillin and clindamycin 4 week	Amoxicillin-clavulanic acid iv 2 week then oral amoxicillin 3 grams/day oral, plan life long
Surgical treatment	Excision and grafting	Excision and iliac-femoral bypass graft	Excision and grafting	Excision and grafting	Excision and grafting	Excision and grafting	Stent-graft and bypass surgery	Stent-graft and bypass surgery
Outcome	Recovery	Recovery	Recovery	Recovery	Recovery	Recovery	Recovery	Recovery

* Tissue culture from an infected aneurysm or site of infection; ** Cozijnsen et al GBS involved thoracoabdominal aorta

However, few case reports including ours confirmed possibilities of atypical manifestation, for example, afebrile with abdominal discomfort in the current report. It is, therefore, challenging for physician since the symptoms can be varied depending on the type of organism, duration of infection and location of an aneurysm. Moreover, the classic manifestations of an infected abdominal aneurysm; specifically abdominal pain, fever, and pulsating abdominal mass, may not fully present altogether on the initial visit/ presentation[6]. Therefore, a complete history and physical examination from a doctor with a high level of awareness to the disease are able to make an early diagnosis resulting in a good clinical outcome.

Early diagnosis of infected AAA is vital in terms of better prognosis and treatment. Since clinical presentation alone may not be able to achieve the diagnosis, laboratory investigations and imagings significantly provide an important role. Laboratory results those might be useful are elevated leukocytosis, elevated erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), and positive hemoculture. Concerning the imaging techniques, CT scan is the investigation of choice due to its availability and its ability to differentiate other aortic emergencies (e.g. dissections). The presence of peri-aortic infiltration with soft tissue collections and gas is highly suggestive of an infective aortic aneurysm. Magnetic resonance imaging (MRI) is an alternative but may not be appropriate, concerning the emergency presentation of the disease, limited by a long waiting time until investigation[16].

In general (generally), diagnosis of invasive GBS infection can be made from the hemoculture. Nonetheless, literature review demonstrates that patients with infected abdominal aortic aneurysm are at the highest possibility of being diagnosed by tissue culture from the suspicious area of infection. Complete thorough laboratory investigation should be performed before administration of antibiotics.

The definite diagnosis of GBS can be achieved from culture on sheep blood agar showing catalase negative, narrow zone beta-hemolysis. Another important test is the CAMP test; the acronym of authors who firstly described-Christie, Atkinson, Munch, and Peterson. The CAMP factor is produced by GBS and acts synergistically with the staphylococcal beta-hemolysin inducing enhanced hemolysis of sheep erythrocytes[17] (Fig. 2A). However, the false positive CAMP test can be found in other pathogens including the group A streptococcus (GAS) and *Listeria* spp. To

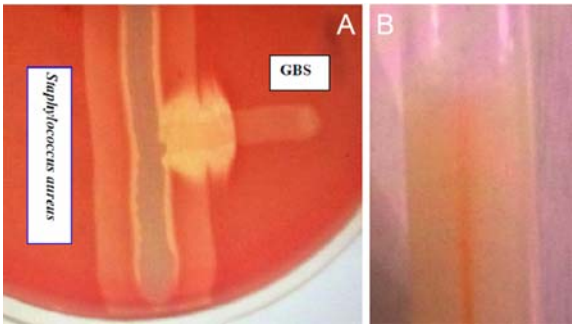

Fig. 2 Characteristics of GBS culture demonstrating positive CAMP test (A) and appearance of orange pigment in Islam's agar (B).

differentiate GBS from these pathogens, GAS colonies commonly demonstrates a wide beta-hemolysis zone and positive Pyrrolidonyl arylamidase (PYR) test whereas GBS colonies are narrow zone beta-hemolytic and PYR test negative. In contrast, *Listeria* spp. is a gram-positive bacillus, catalase positive bacteria. Furthermore, another very useful rapid test in identifying GBS is to culture in the Islam's agar. GBS produces an orange pigment which will show on the Islam's agar (Fig. 2B). The test is highly sensitive and specific[18].

An infected abdominal aortic aneurysm is associated with a high morbidity and mortality rate. The principle of treatment is control or eradication of infection followed by an establishment of arterial reconstruction. The combination of proper antimicrobial agents and adequate surgical management is crucial to earning the best clinical outcomes. Surgical management should be tailored made appropriate to each individual. In general, penicillin (penicillin G, ampicillin) is the antibiotic of choice for treatment of GBS infection. In penicillin allergies, vancomycin is the initially recommended substitute. Erythromycin or clindamycin should not be used in penicillin allergies, due to increasing resistance, unless sensitivity tests show susceptibility to these agents. Combined gentamicin and penicillin is used, for synergy, in patients with life-threatening GBS infections. However, if an aminoglycoside is to be added to penicillin or ampicillin in treating GBS, a sensitivity test for aminoglycoside is recommended since synergy does not occur if the organism is not sensitive to aminoglycosides[19-21].

There is no consensus for the duration of antibiotic therapy. Generally, antibiotics can be discontinued if there is no clinical, laboratory, or imaging evidence of ongoing sepsis. However, some authors

recommend long durations and even life-long antibiotic therapy, especially in the prosthetic reconstruction[3,6]. The hallmark for surgical treatment of an infected aneurysm includes: control of bleeding, debridement of non-viable and infected tissue, and arterial reconstruction[6].

Our patient received amoxicillin/clavulanic acid intravenously for 2 weeks with good clinical response. After the subsiding of abdominal pain and improvement of other clinical signs, life-long oral amoxicillin 3 grams/day was given. We followed-up this patient for 20 months without any recurrent infections and good clinical response.

Conclusion

In conclusion, the authors report a very rare case of infected infrarenal abdominal aortic aneurysm due to GBS infection. This is the first case report in Thailand.

What is already known on this topic ?

Group B streptococcus (GBS) is usually harmless in healthy human beings. It is capable of colonizing in normal gastrointestinal and genitourinary tracts, called asymptomatic carriers. However, if the host becomes immunocompromised, it could cause various serious infections which could be life-threatening. Common infections found are endocarditis, meningitis, and bacteremia without a primary source of infection. In invasive disease, the diagnosis is usually made through positive hemoculture. GBS is typically susceptible to penicillins.

What this study adds ?

A mycotic aneurysm or infected aneurysm of the aorta is quite uncommon. Moreover, group B streptococcus infection is an extremely rare cause of this. This case report adds to the literature of the infected abdominal aortic aneurysm caused by GBS in the elderly. Our case had a unique presentation of abdominal pain without fever. Diagnosis is difficult and based on CT scan. Therefore, an initial high index of suspicion in doctors is vital in patients with chronic abdominal pain and a pulsatile mass. Doctors should conduct immediate further inventions to obtain an early diagnosis and treatment, which would lead to better prognosis. After conducting a literature review, tissue culture from the suspicious area of infection exhibits the highest yield for diagnosis. Complete laboratory investigations should be obtained before administering antibiotics. The management of these conditions is difficult. The combined medical-surgical approach with early empirical broad-spectrum antibiotics given seems to offer the best results.

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Potential conflicts of interest

None.

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การติดเชื้อหลอดเลือดโป่งพองของเส้นเลือดแดงเอออร์ตาในช่องท้องจากเชื้อสเตร็ปโตค็อกคัสกรุ๊ป บี: รายงานผู้ป่วยและการทบทวนวรรณกรรม

ประสิทธิ์ อุพาพรรณ

รายงานผู้ป่วยที่พบน้อยมากถึงการติดเชื้อแบคทีเรียสเตร็ปโตค็อกคัสกรุ๊ป บี ที่เส้นเลือดแดงใหญ่เอออร์ตาในช่องท้องในผู้ป่วยชายอายุ 66 ปี ที่ไม่มีไข้ มีโรคประจำตัวความดันโลหิตสูงและสูบบุหรี่ มาพบแพทย์ด้วยอาการปวดอืดแน่นท้อง ตรวจร่างกายพบก้อนลักษณะคล้ายซีพจรเต้นบริเวณส่วนกลางของช่องท้องขนาดห้าเซนติเมตร ผลตรวจเอ็กซเรย์คอมพิวเตอร์ช่องท้องพบเส้นเลือดโป่งพอง บริเวณเส้นเลือดแดงใหญ่เอออร์ตาในช่องท้องส่วนที่อยู่ใต้ต่อเส้นเลือดแดงที่ไตขนาดประมาณห้าเซนติเมตร ได้รับการตรวจหัวใจด้วยคลื่นเสียงความถี่สูงผ่านผนังทรวงอกไม่พบความผิดปกติที่เยื่อบุหัวใจหรือการติดเชื้อที่ลิ้นหัวใจ ได้รับการผ่าตัดซ่อมแซมเส้นเลือดโป่งพองที่เส้นเลือดแดงใหญ่เอออร์ตาในช่องท้องและได้รับยาอะม็อกซีซิลลินคลาวูลานิกเอซิดทางหลอดเลือดดำ ผลเพาะเชื้อจากชิ้นส่วนผนังเส้นเลือดโป่งพองและต่อมน้ำเหลือง บริเวณเส้นเลือดแดงใหญ่เอออร์ตาพบเชื้อแบคทีเรียสเตร็ปโตค็อกคัสกรุ๊ป บี ให้การรักษาต่อด้วยยาอะม็อกซีซิลลินคลาวูลานิกเอซิดทางหลอดเลือดดำรวมสิบสี่วันและได้เปลี่ยนเป็นยาอะม็อกซีซิลลินรับประทานสามกรัมต่อวัน ปัจจุบันได้ติดตามอาการผู้ป่วยรวมยี่สิบเดือนอาการผู้ป่วยปกติดี ไม่พบการติดเชื้อซ้ำหรือภาวะแทรกซ้อนภายหลังการผ่าตัด รายงานนี้จัดเป็นรายงานผู้ป่วยรายแรกในประเทศไทย ที่ติดเชื้อแบคทีเรียสเตร็ปโตค็อกคัสกรุ๊ป บี ที่ตำแหน่งเส้นเลือดแดงเอออร์ตาในช่องท้อง