·临床研究·

主动脉窦瘤的超声特点与漏诊分析

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摘　要：【目的】总结主动脉窦瘤(SVA)的超声特点，分析漏诊原因，探讨SVA的超声诊断技巧，提高诊断率。【方法】回顾性分析2014年1月至2022年3月期间在中山大学附属第一医院行外科手术的52例SVA患者的超声心动图特征及手术资料，按改良Sakakibara分型分为5型。【结果】52例SVA患者中，男性32例，女性20例，年龄18~66(36.1±11.6)岁。其中44例窦瘤起自主动脉右冠窦，8例起自无冠窦，未见起自左冠窦的病例。Ⅰ型、Ⅱ型和Ⅲv型为膨入右心室的窦瘤共35例，32例(91.4%)合并室间隔缺损(VSD)，Ⅲa型、Ⅳ型和Ⅴ型为膨入右心房或其他部位的窦瘤共17例，仅2例(17.6%)合并VSD。各型SVA均常合并主动脉瓣病变，因主动脉瓣病变情况较严重需要外科手术换瓣或成形者共27例(51.9%)。52例患者共有4例漏诊SVA，漏诊率为7.7%；8例漏诊VSD，漏诊率达23.5%，漏诊者多为Ⅰ型SVA合并干下型VSD；各型SVA中均有合并感染性心内膜炎(IE)患者，共19例，其中有2例漏诊，漏诊率为10.5%。【结论】SVA声像图具有多样性和复杂性。膨入右心房的SVA较少合并VSD，而膨入右心室的SVA绝大多数合并VSD，但诊断具有挑战性，且SVA还易合并主动脉瓣病变和IE，进一步加大诊断难度。超声检查过程中必须提高警惕，灵活运用多个标准和非标准切面，减少漏诊，提高诊断率。

关键词：主动脉窦瘤；超声心动描记术；漏诊
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Echocardiographic diagnosis of sinus of Valsalva aneurysm

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Abstract:【Objective】To summarize the echocardiographic features of sinus of Valsalva aneurysm (SVA), analyze the causes of missed diagnosis, thus explore the diagnostic skills and improve the diagnostic accuracy for SVA.【Methods】The echocardiographic features and clinical data of 52 SVA patients who underwent surgery in the First Affiliated Hospital of Sun Yat-sen University from January 2014 to March 2022 were retrospectively reviewed. The patients were divided into 5 types according to modified Sakakibara classification system.【Results】There were 32 male and 20 female patients with their age of 18~66 (36.1±11.6) years. Of the 52 aneurysms, 44 originated from the right coronary sinus (RCS), 8 from noncoronary sinus (NCS) and none from left coronary sinus (LCS). Among the 35 SVAs protruding into the right ventricle, including type Ⅰ, type Ⅱ and type Ⅲv, 32 (91.4%) were associated with ventricular septal defect (VSD). There were 2 (17.6%) associated with VSD among the 17 SVAs protruding into the right atrium or other sites of the heart, including type Ⅲa, type Ⅳ and type Ⅴ. SVA was frequently associated with aortic valve disease, 27 cases (51.9%) of which needed surgical valve replacement or valvoplasty. SVA was missed in 4 patients and VSD in 8, with the misdiagnosis rates of

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7.7% and 23.5%, respectively. The most commonly missed VSD diagnosis was subarterial VSD with type I SVA. Of the 19 SVAs associated with infective endocarditis (IE), 2 were missed, with the misdiagnosis rate of 10.5%.【Conclusion】The ultrasound images of SVA are diverse and complex. SVA protruding into the right atrium is rarely associated with VSD, while SVA protruding into the right ventricle is frequently associated with VSD. SVA is also prone to be associated with aortic valve disease and IE, which makes the diagnosis more challenging. Therefore, during ultrasound examination, we must vigilantly and flexibly make use of the multiple scan slices so as to decrease the rate of missed diagnosis and improve the diagnostic accuracy for SVA.

Key words: sinus of Valsalva aneurysm (SVA); echocardiography; missed diagnosis

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主动脉窦瘤(sinus of Valsalva aneurysm,SVA)是一种少见的心血管疾病,表现为局部窦壁向外呈瘤样膨出,常发生于右冠窦及无冠窦,多为先天性,且常合并先天性室间隔缺损(ventricular septal defect,VSD)[1]。SVA一旦破裂,会引起显著的血流动力学变化,若不及时诊断治疗,易引起心力衰竭等严重后果,平均存活时间为1~2年,预后较差;若能及时手术治疗,可显著改善患者临床症状,延长生存期,预后良好[2];因此及时正确的诊断对SVA患者至关重要。虽然计算机断层摄影术和磁共振成像技术已广泛用于心血管疾病的诊断,但超声心动图仍是诊断SVA的首选影像学检查方法[3]。本研究回顾性分析中山大学附属第一医院近8年来52例行外科手术SVA患者的超声特点,旨在探讨超声心动图漏诊原因及诊断技巧,提高诊断准确性。

1 材料与方法

1.1 研究对象

2014年1月至2022年3月于中山大学附属第一医院超声心动图或/和心脏外科手术诊断的SVA患者共63例,排除未手术患者11例,本研究共纳入经外科手术证实的SVA患者52例。本研究的实验方案及相关细节依据《赫尔辛基宣言》,并经中山大学附属第一医院医学伦理委员会批准。纳入标准:年龄大于18岁;心脏外科手术证实为SVA的患者;术前超声心动图检查考虑为SVA的患者。排除标准:超声心动图检查考虑SVA,但心脏外科手术证实非SVA的患者。

1.2 仪器与方法

1.2.1 仪器 采用Philips iE33、Philips EPIQ 7C、GE vivid7、GE vividE95超声诊断仪,使用成人经胸二维相控阵探头,频率为1.0~5.0 MHz或1.7~3.4 MHz。

1.2.2 临床资料 回顾SVA患者的临床资料,记录明确诊断的SVA患者的外科手术资料,包括手术方式、手术记录和术后诊断。

1.2.3 超声心动图图像采集及数据测量 所有患者术前均行超声心动图检查,记录常规参数及SVA的相关数据,包括心脏各腔室大小和左心室射血分数;SVA的起源、膨入部位、大小、窦壁回声、破口的数量和大小、有无异常回声和流出道梗阻;主动脉瓣形态和功能;有无合并其他先天性畸形;彩色多普勒血流显像观察破口处血流的特点、连续多普勒记录相应血流动力学情况。

1.2.4 SVA分型 按改良Sakakibara分型[4]法分为5型。Ⅰ型:窦瘤在近肺动脉瓣处膨入右心室;Ⅱ型:窦瘤在室上嵴及嵴下部位膨入右心室;Ⅲv型:窦瘤在三尖瓣环或紧邻三尖瓣环处膨入右心室;Ⅲa型:窦瘤在三尖瓣环或紧邻三尖瓣环处膨入右心房;Ⅳ型:窦瘤膨入右心房;Ⅴ型:窦瘤膨入肺动脉、左心房、左心室或其他部位。

1.3 统计学分析

使用SPSS 25.0统计软件对数据进行统计分析,正态分布的连续性变量用其均数±标准差表示,非正态分布的连续性变量,用中位数(下四分位数~上四分位数)表示,计数资料以例(%)表示。

2 结 果

2.1 基本情况

本研究共纳入52例患者,男性32例,女性20例,年龄范围18至66岁,平均36.1±11.6岁,其中右冠窦瘤44例(5例未破裂),无冠窦瘤8例(均破裂),未发现左冠窦瘤病例。主动脉窦瘤膨入心腔情况见表1。膨入右心室和右心房的SVA共50例,占96%,其中膨入右心室的SVA共35例,5例未破裂,膨入右心房的SVA有15例,均破裂。

2.2 超声心动图基本特征

2.2.1 直接征象 超声心动图检查可见主动脉局部窦壁向外呈瘤样膨出,于不同方向膨入邻近结构(图1)。多数窦瘤壁可见破口,彩色多普勒血流显像可见破口处舒张期为主的连续性高速左向右分流信号,连续多普勒频谱表现为舒张期分流速度逐渐增强或舒张中期达峰(图2)。右冠窦瘤破入左心室是特殊类型的SVA,表现为起自主动脉根部的囊袋样结构,膨入左室流出道,瘤体舒张期膨大、收缩期塌陷,彩色多普勒血流显像仅可见舒张期的分流信号,连续多普勒频谱与主动脉瓣反流频谱类似。

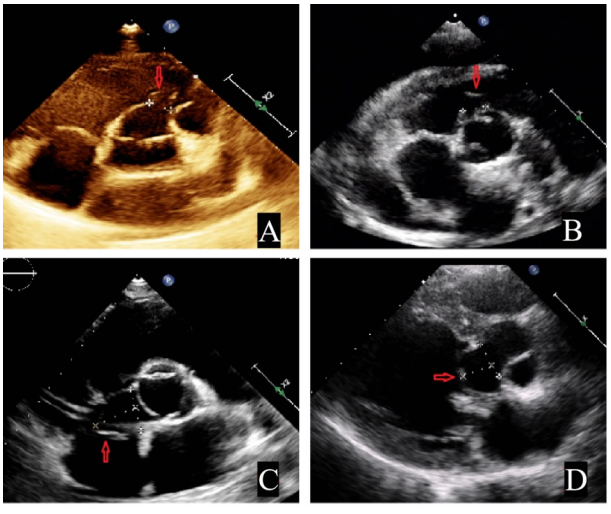
2.2.2 间接征象 破入右心室的SVA心脏继发改变主要表现为主动脉及肺动脉增宽、左心房及左心室增大,破入右心

表1　主动脉窦瘤膨入心腔情况

Table 1　Origin and Termination of SVAs

Origin	Termination			Cases (%)
	RV	RA	Others	
RCS	34	9	1 [1)]	84.6
NCS	1	6	1 [2)]	15.4
LCS	0	0	0	0
Tota/l%	35(67.3)	15(28.8)	2(3.8)	100

RV: right ventricle; RA: right atrium; RCS: right coronary sinus; NCS: noncoronary sinus; LCS: left coronary sinus; [1)]: right-SVA rupture into left ventricle; [2)]: non-SVA rupture outside the heart cavity and form a giant pseudoaneurysm.



A showed type Ⅰ SVA protruding into right ventricle just beneath pulmonary valve. B showed type Ⅱ SVA expending into or just beneath crista supraventricular is of right ventricle. C showed type Ⅲa SVA protruding into the right atrium adjacent to tricuspid annulus. D showed type Ⅴ SVA distention into the left ventricular outflow tract. SVAs were indicated by the red arrow.

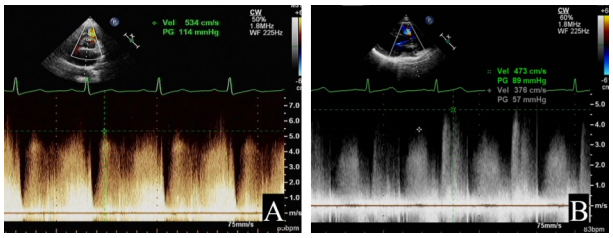
图1　不同类型SVA二维声像图

Fig. 1　Two-dimensional sonographic images of different types of SVA

房的SVA尚出现右心房右心室增大(表2)。51例SVA患者的左心室射血分数在正常范围，仅1例右冠窦瘤膨入右心室的患者左心室射血分数减低至34%。

2.3　主动脉窦瘤的类型分析

按照Sakakibara改良分型法[4]将患者分成5型。各类型具体分布情况见表3。Ⅰ型SVA为最常见的类型，共26例，其中23例破裂；Ⅱ型共7例，其中5例破裂；Ⅲ型共16例，均破裂，以Ⅲa型(14例)多见；Ⅳ型和Ⅴ型病例数较少，仅3例，窦瘤均破裂。



A was a typical continuous high-speed SVA shunt with gradually increasing velocity in diastole. B showed the continuous high-speed SVA shunt, in which the diastolic shunt reached its peak in the middle diastole.

图2　SVA分流的连续多普勒频谱图像

Fig. 2　Continuous Doppler spectrum image of SVA shunt

表2　SVA破入右心室与右心房常规超声参数情况

Table2　Echocardiographic parameters of SVA with termination to RV and RA　[$M(P_{25}\sim P_{75})$]

Echocardiographic parameters	Termination	
	RV (n=30)	RA (n=15)
SoVAo /mm	36 (33~38)	36 (33~37)
LA /mm	35 (32~44)	38 (33~45)
LVIDd /mm	56 (50~64)	56 (54~59)
RA length /mm	45 (40~50)	56 (51~59)
RA transverse diameter /mm	36 (32~39)	46 (41~54)
RV /mm	22 (20~25)	25 (22~30)
MPA /mm	26 (23~30)	26 (23~29)
LVEF /%	67 (63~71)	67 (61~71)

SoVAo: sinus of Valsalva; LA: left atrium; LVIDd: left ventricular internal dimension diastole; RA: right atrium; RV: right ventricle; MPA: main pulmonary artery; LVEF: left ventricular ejection fraction.

2.4　合并心脏病变分析

SVA最常见的合并畸形是先天性VSD，本研究中65.4%的患者合并VSD。分析发现，是否合并VSD与SVA位置有密切关系，Ⅰ型、Ⅱ型以及Ⅲv型为膨入右心室的SVA，有32例(91.4%)合并VSD，其中23例为干下型，4例嵴内型，5例膜周型；Ⅲa型、Ⅳ型和Ⅴ型为膨入右心房或其他部位的SVA，仅2例(17.6%)合并VSD，分别为膜周型和隔瓣下型。

主动脉瓣病变也是SVA常见的心脏并发症(图3)。本研究共29例(55.8%)患者出现主动脉瓣形态改变，主要包括瓣叶增厚、钙化、脱垂、挛缩和穿孔；34例(65.4%)患者出现主动脉瓣反流(aortic valve regurgitation，AR)；另有1例主动脉瓣狭窄、2例主动脉瓣下隔膜与1例先天性二叶式主动脉瓣。因主动脉瓣病变严重需外科换瓣或成形者共27例，手术率高达51.9%。膨入右心室的SVA有27例(77.1%)出

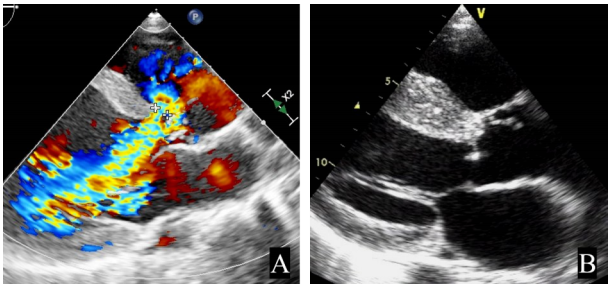
表3 各型SVA合并心脏病变情况

Table 3 Cardiovascular lesions associated with different types of SVA

Type	Cases (n)	Male (n)	Associated cardiovascular lesions (n)				
			VSD	IE	AR	RVOTO	Others
Ⅰ	26	13	24	8	19	1	3 [1)]
Ⅱ	7	5	6	6	6	2	0
Ⅲ	16	12	4	4	6	0	1 [2)]
Ⅲv	2	1	2	1	2	0	0
Ⅲa	14	11	2	3	4	0	1 [2)]
Ⅳ	1	1	0	0	1	0	0
Ⅴ	2	1	0	1	2	0	1 [3)]
Total	52	32	34	19	34	3	5

VSD: ventricular septal defect; IE: infective endocarditis; AR, aortic valve regurgitation; RVOTO, right ventricular outflow tract obstruction; [1)]: 2 cases of discrete subaortic membrane and 1 case of patent ductus arteriosus; [2)]: tricuspid valvular malformation; [3)]:congenital bicuspid aortic valve.

现AR,其中需外科手术者共20例(57.1%);膨入右心房的SVA有5例(33.3%)出现AR,5例均需外科手术治疗;2例Ⅴ型SVA患者均合并严重的AR,且均需外科手术治疗。

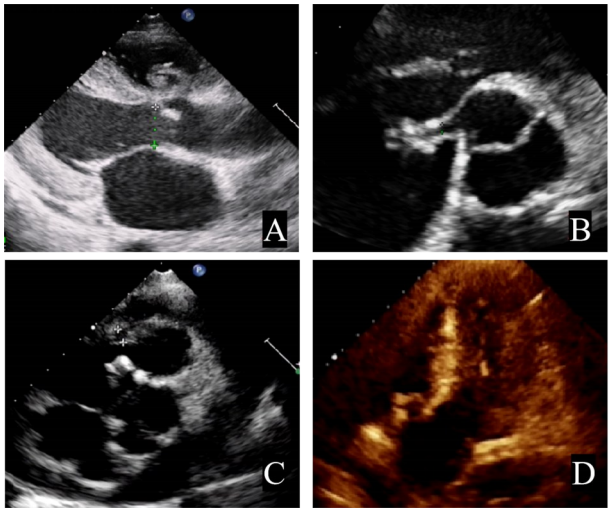


A was a Color Doppler image showing severe aortic regurgitation. B was a two-dimensional image showing the subaortic septum.

图3 SVA合并主动脉瓣病变胸骨旁左心室长轴声像图

Fig. 3 Parasternal long axis view of left ventricle in SVA with aortic valve disease

52例SVA患者中有19例(36.5%)合并感染性心内膜炎(infective endocarditis, IE)。外科术中探查可在窦瘤内、破口处、VSD右室面、主动脉瓣及三尖瓣上见到赘生物,亦可见主动脉瓣叶穿孔,未见主动脉根部脓肿及瘘管形成。膨入右心室的SVA有15例(42.8%)合并IE;膨入右心房的SVA有3例(20%)合并IE;Ⅴ型SVA患者中有1例合并IE。合并IE的病例均可见窦瘤壁增厚、厚薄不均或局部有强回声钙化(图4)。



A was a long-axis view of the left ventricle, showing the right coronary sinus aneurysm expanding into the right ventricle, and the aneurysm wall and aortic valve were significantly thickened. B was short-axis view of the aortic root, showing the right coronary sinus aneurysm expanding into the right atrium, and the aneurysm wall was significantly thickened and calcified. C and D were the short-axis view of aortic root and the apical five-chamber view, respectively. Both showed the right coronary sinus aneurysm expanding into the right ventricle, with obvious thickening and calcification of the aneurysm wall.

图4 SVA合并IE声像图

Fig. 4 Ultrasound images of SVA combined with IE

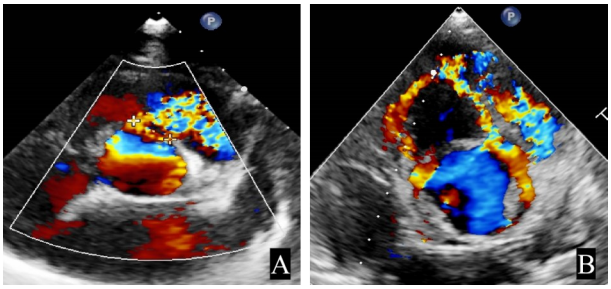
其他合并心脏病变情况:有3例膨入右心室的窦瘤造成右室流出道梗阻;1例Ⅰ型窦瘤合并动脉导管未闭;Ⅲa型患者中因三尖瓣反流严重而需三尖瓣成形者共6例(42.8%),其中有1例为三尖瓣前瓣与隔瓣交界处部分瓣叶缺损的先天性畸形。

2.5 超声心动图诊断结果与手术结果比较

52例SVA患者中,术前超声心动图检查明确诊断者共48例,漏诊4例,漏诊率为7.7%。超声心动图诊断的VSD共26例,漏诊8例,漏诊率为23.5%。在漏诊的8例室缺中,5例为Ⅰ型SVA合并干下型VSD(图5);2例为Ⅱ型SVA,分别合并干下型和嵴内型VSD;1例为Ⅲa型SVA合并膜周型VSD。另有3例VSD断口大小超声测量误差较大,将大VSD诊断为小VSD(图5),误差值7mm~20mm不等。19例合并IE患者中有2例漏诊,漏诊率为10.5%。其他合并畸形,如3例右室流出道梗阻、2例主动脉瓣下隔膜、1例二叶式主动脉瓣与1例动脉导管未闭,超声心动图均给出了准确诊断。

3 讨论

主动脉窦瘤是一种少见的心血管疾病,表现为主动脉窦壁局部向外膨出并破入附近腔室或心包,从而引起一系



A showed that SVA shunt completely occluded VSD shunt, and it was challenging to judge whether there was combined with a VSD. B showed that SVA embedded in VSD, which led to obvious thinning of VSD shunt.

图5　SVA合并VSD胸骨旁短轴切面彩色多普勒图像

Fig. 5　Parasternal short-axis view with Color Doppler image of SVA combined with VSD

列严重的血流动力学变化。该疾病男性居多，亚洲人群发病率为西方人群的5倍[4-5]。SVA多为先天性，其形成原因主要为胚胎发育过程中主动脉壁中层弹力纤维与主动脉瓣环融合不良，导致局部窦壁弹性纤维层缺乏，在高压力的持续作用下，薄弱部位逐渐向外膨出形成SVA甚至破裂[6-7]。最常见的SVA是右冠窦受累(65%~85%)，其次是无冠状窦(10%~30%)，左冠窦几乎不受累，这一现象与胚胎起源有关[8]。不断扩大的窦瘤通过压迫或直接破裂到邻近的腔室而引起胸闷等临床症状，50%至70%的患者临床表现为渐进性，多于中青年时期发病[9]。窦瘤一旦发生破裂，未治疗的患者平均存活期仅为1~2年，如及时手术则预后良好，因此该病需尽早诊断，及时治疗[9]。

超声心动图是SVA的首选检查方法[3]，具有较高的敏感性和准确性[2]。超声检查可清晰显示主动脉窦壁局部向外呈“风袋”样膨出，彩色多普勒血流显像可见破口处舒张期为主的高速连续性分流，同时对心脏功能、并发畸形和继发心脏改变进行评价和诊断[7]。95%以上的SVA膨入/破入右心房和右心室[4]，其中破入右心室的SVA心脏继发改变为主动脉及肺动脉增宽、左心房及左心室增大；破入右心房的SVA尚出现右心房右心室增大。绝大多数SVA患者的左心室射血分数在正常范围。本研究共纳入52例SVA患者，术前经胸超声心动图明确诊断48例，且对窦瘤起源、大小、膨入部位、破口大小的判断均与术中所见基本一致，诊断准确率为92.3%。回顾漏诊的4例SVA的超声图像，其中2例漏诊原因是窦瘤壁菲薄，二维图像显示欠佳且没有连接心电图，误诊为VSD；另2例漏诊，是对罕见SVA类型认识不足，分别是右冠窦瘤破入左室流出道和无冠窦瘤破入心腔外形成假性动脉瘤并瘤内血栓。

SVA常合并先天性心脏畸形，其中约60%~70%的SVA患者合并VSD[10]。本组病例中，Ⅰ型、Ⅱ型以及Ⅲv型为膨入右心室的SVA，合并VSD的比例高达91.4%，并且VSD位置多位于SVA正下方。推测两者可能具有相似的先天发育异常的病理基础，亦可能与VSD的高速血流对邻近主动脉窦的虹吸作用有关[11]。术前明确SVA是否合并VSD直接影响手术方案的制定，即使是合并小的VSD，外科医生对VSD及时的修补对保留主动脉瓣和肺动脉瓣的功能至关重要[12-13]。然而，在SVA患者中识别是否合并VSD通常是较为困难的[14]。本组病例中有8例漏诊了VSD，同时有3例对VSD大小的判断也存在较大误差。回顾分析发现漏诊的主要原因是SVA部分或完全遮挡VSD，使得VSD的分流明显减小，甚至无分流。我们常在大动脉短轴切面观察VSD的位置和大小，然而由于SVA的遮挡，大动脉短轴切面不再是判断是否存在VSD的最佳切面。因为SVA位于主动脉瓣环的上方，而VSD位于瓣环的下方，此时，应主要依据显示上下解剖方位关系的切面判断是否合并VSD，如左心室长轴切面、心尖五腔切面或右室流出道切面等，配合探头扫查角度的调整，观察室间隔和主动脉瓣环之间是否有回声中断[12]。彩色多普勒血流显像在SVA合并VSD诊断中也发挥重要作用，首先VSD与SVA二者发生的时相不同，VSD是收缩期分流，SVA则是以舒张期为主的连续性分流；其次分流起始位置不同，VSD的分流始于室间隔的平面，而SVA的分流则多数出自其顶端的破口。虽然二者的分流频谱存在明显不同，但SVA的连续分流频谱常掩盖VSD的收缩期频谱[6]，此时需综合分析仔细判断。测量VSD大小时，也应考虑SVA的遮挡，VSD的分流束会变小甚至无分流，笔者建议二维超声直接测量VSD的断口，不要过度依赖分流束宽度。

SVA还易合并主动脉瓣病变，包括主动脉瓣增厚、钙化、脱垂和反流等[15]。本研究中高达65.4%的患者合并AR，亦有主动脉瓣狭窄、瓣下隔膜、二叶式主动脉瓣等情况，需同期行主动脉瓣置换术或主动脉瓣成形术的比例高达51.9%。本研究发现，主动脉瓣病变情况与SVA的膨入部位相关，其中膨入右心室的SVA合并主动脉瓣病变比例及手术率均高于膨入右心房的SVA。主动脉瓣病变也进一步增加了诊断SVA的难度，分析其原因主要有两点：第一，严重的AR和SVA破裂都会在降主动脉内出现舒张期反向血流频谱；第二，VSD合并AR时，连续多普勒会显示类似SVA的双期频谱，收缩期是VSD的频谱，舒张期是主动脉瓣反流的频谱。回顾分析发现，SVA和AR的频谱在舒张期有较大差异，AR的频谱呈现典型的舒张早期达峰，之后逐渐减弱，而SVA的频谱则表现为舒张期逐渐增强或舒张期中期达峰[8, 16]。同时结合二维图像和彩色多普勒成像，可降低两者鉴别难度。

SVA亦是诱发IE的常见因素。本组病例中，高达36.5%的SVA患者合并IE，其中在膨入右心室的SVA或合并VSD的病例中，该比例进一步增加到40%以上，明显高于单纯VSD合并IE的比例[17]。这为检查者敲响警钟，对于SVA的患者，尤其是膨入右心室或合并VSD的患者，要仔细辨别有无合并IE，特别注意窦瘤内、破口处、VSD的断端、湍流束的下游及邻近的瓣叶等位置，建议局部放大二维图像，以便发现细小的赘生物。本研究回顾超声图像发现，未合并IE的患者，窦瘤壁通常较薄且回声均匀或厚度与正常瓣叶相当，而窦瘤壁的异常回声表现仅出现在合并IE的病例中，尤其对于窦瘤壁增厚、厚薄不均或局部有强回声钙化

者,需高度警惕有无合并IE。

本研究具有一定局限性,为单中心回顾性研究,纳入病例数有限,要进一步总结主动脉窦瘤的特点及漏诊原因,需要多中心、大样本数据予以进一步证实。

综上所述,SVA超声影像具有多样性和复杂性,常合并VSD、主动脉瓣病变和IE,且病变间相互影响,给超声心动图的准确诊断带来极大挑战。检查过程中,对于膨入右心室的SVA,密切注意有无合并VSD,同时仔细观察主动脉瓣病变情况;对于窦瘤壁不均匀增厚或有强回声钙化时,要考虑IE的可能;应在标准切面的基础上,结合非标准切面,局部放大图像,综合多普勒信息,准确诊断SVA并判断有无合并症,以期为外科手术决策提供可靠的诊断信息和依据。

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