Kommerell Diverticulum: A Five Year Incidence and Morphology Study Using Computed Tomography

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The objective is to study the incidence and morphology of aberrant subclavian arteries (ASCA) and Kommerell diverticulum (KD) to resolve long-standing controversies.

We retrospectively reviewed 30898 chest CT scans/patients over a five-year period and found 195 (0.63%) patients with ASCA. Excluding 4 non-enhanced scans, 191 patients were included for analysis. The branching pattern (right arch with aberrant left subclavian artery (ALSCA) or left arch with aberrant right subclavian artery (ARSCA)), the diameters of ASCA at ostium (ASCA-O) and at midline (ASCA-M) were recorded. ASCA-O/ASCA-M ratio (O/M ratio) equal to or larger than 1.5 was defined as KD. We further analyzed the correlation among several factors including age, sex, ALSCA/ARSCA, ASCA-O, ASCA-M, O/M ratio, and presence of KD.

All 191 ASCA coursed behind the esophagus. Most instances of KD were found in older ASCA patients. The growth rate of ASCA-O (0.12 mm/year) was 1.5 times faster than ASCA-M (0.08 mm/year). ALSCA patients had a higher incidence of KD than ARSCA patients. ALSCA patients older than 50 years had 100% (4/4) incidence of KD formation.

We concluded that (1) all ASCA course behind the esophagus, (2) KD formation is a combination of congenital and acquired processes, and (3) ALSCA and age are the two risk factors for KD formation.

Kommerell diverticulum (KD) is an aneurysmal dilatation at the origin of an aberrant subclavian artery [1, 2]. Initially, Kommerell described a KD case with left aortic arch and aberrant right subclavian artery (ARSCA) in 1936 [1]. In the following decades, KD was also found in patients with right aortic arch and aberrant left subclavian artery (ALSCA) [3-5]. Some articles focusing on the image appearance [3, 6, 7], surgery [8] and incidence [3, 5] have been published. However, several long-standing controversial issues have not been resolved. First, KD is traditionally considered to be a congenital disease [1, 2, 9, 10]. However, several authors think atherosclerosis plays a major role in the formation of KD [11-13]. Furthermore, if KD formation is related to age, what is the annual growth rate? Is the growth rate in the ostium different from that in the more distal segment of the aberrant subclavian artery (ASCA)? Second, is the incidence of KD really higher in ALSCA patients than ARSCA patients, as proposed by some investigators [2, 3, 5] based on their clinical experience? Do these two mirror image aortic arch branching patterns (ARSCA vs. ALSCA) have different incidences of KD? Third, it is frequently stated that ASCA course behind the esophagus in 80% of cases, between the trachea and esophagus in 15% and in front of the trachea in 5% [13]. However, this statement has long been challenged [6, 7]. To date, the three abovementioned questions have not been answered because the
published articles are mostly case reports [8, 9, 11] or case series with limited numbers of patients [3-5]. We retrospectively reviewed the five-year image database of a 1,500-bed tertiary referral center to obtain a large cross-sectional result in order to answer these controversial questions.

**MATERIALS AND METHODS**

**Patient enrollment**

We retrospectively reviewed all chest CT scans performed in a 1,500-bed tertiary referral medical center.

**Figure 1.** A 13-year-old asymptomatic girl had suspected vascular ring demonstrated by echocardiography for anatomy clarification. These serial axial images demonstrate the measurements used in this study. **a.** Axial CT image at the level of the thoracic inlet shows the major branches of the aortic arch. The aberrant right subclavian artery (ARSCA) is identified. (E: esophagus, LCCA: left common carotid artery, LSCA: left subclavian artery, RCCA: right common carotid artery, T: trachea). **b.** Axial CT image slightly more caudal than figure 1a shows the ASCA-M measurement. ASCA-M (double arrowhead) was defined as the diameter of the aberrant subclavian artery at the midline. (E: esophagus, LCCA: left common carotid artery, LSCA: left subclavian artery, RCCA: right common carotid artery, T: trachea). **c.** Axial CT image slightly more caudal than figure 1b shows that the aberrant right subclavian artery (double arrowhead) is located behind both the trachea (T) and the esophagus (E). The measurement of ASCA-O, which was defined as the diameter at the ostium of the aberrant subclavian artery, is indicated by the double arrowhead. With the measurements of ASCA-O and ASCA-M, the O/M ratio could be calculated. The definition of Kommerell diverticulum in this study was an O/M ratio larger than or equal to 1.5. (Arch: Aortic arch, E: esophagus, T: trachea)
center from January 2005 to December 2009 and enrolled all patients with ASCA (either ARSCA or ALSCA) evidenced by the imagery. Patients who underwent more than one CT scan were included only for the earliest scan. Because the measurement of the vessel size and evaluation of degenerative change required contrast injection, non-enhanced CT scans were excluded. We didn’t review the presenting symptoms because (1) ASCA itself is usually an asymptomatic anomaly, and even if it is symptomatic, the symptoms are usually non-specific and (2) this study focused on the incidence and morphology. These patients were referred for various indications including chest survey, underlying disease evaluation (lung cancer, mediastinal tumor, etc.) or suspected vascular ring. This retrospective study was approved by our institutional review board, which waived the requirement for informed consent.

CT scan

Chest CT studies were performed by using a 64-MDCT scanner (Brilliance 64, Philips, Best, The Netherlands) and a single slice spiral CT scanner (PQ 6000, Picker International Inc., Cleveland, USA). Intravenous contrast medium injection (Omnipaque 350, GE Healthcare, Milwaukee, USA) was injected in all patients with a dose of 100 mL. The parameters of the 64-MDCT scanner were a tube voltage of 120kV, a tube current of 250 mAs/slice, a pitch of 0.797, rotation time of 0.75 second and a collimation of 64*0.625 mm. The parameters of the single slice spiral CT were 120 kV, 200 mA, rotation time of 1 s, and a pitch of 2.0. Contrast medium was injected at a flow rate of 2.0 ml/s and the scan started after a 45-second delay. We obtained serial axial images from the lower neck to the adrenal glands, with slice thickness and index of 5 mm. For pediatric patients with body weight less than 50 kg, 64-MDCT was performed with previously described protocols [14, 15].

Image analysis and measurements

One senior chest radiologists with 10 years of chest CT experience reviewed all chest CT scans on the PACS system using a DICOM viewer (Centricity 6.1, GE Healthcare, Milwaukee, USA) with standard soft tissue window settings (window level/width: 60/450 HU). Patients with ASCA were enrolled into the study. The sex and age were recorded. The pattern (right side aortic arch with ALSCA or left side aortic arch with ARSCA) and the course of ASCA (posterior to the esophagus, between the esophagus and the trachea or anterior to the trachea) were also identified and recorded. The diameter of the ASCA ostium was recorded as ASCA-O (Fig. 1). When the ASCA crosses the midline, the volume-average effect at midline is less significant, so the diameter in such cases was recorded as ASCA-M (Fig. 1). Then, we calculated the ASCA-O/ASCA-M ratio and recorded it as the O/M ratio. Since there is no universal definition of KD, we used the criteria concept proposed by the Society for Vascular Surgery [16]. An O/M ratio equal to or larger than 1.5 was defined as positive for KD (Fig. 2). The degenerative change at the ASCA ostium was also classified into four degrees and recorded. The degrees were defined as: 1 - no degenerative change, 2 - only mural calcifications, 3 - mural thrombus or mural calcification less than 10 mm in thickness, and 4 - mural thrombus or mural calcification equal to or more than 10 mm in thickness (Fig. 3).

Figure 2. A 14-year-old boy with chief complaint of swallowing difficulty. Under the suspicion of vascular ring, he underwent chest CT for evaluation. MDCT confirmed the presence of aberrant right subclavian artery. Volume-rendered 3D image was then reconstructed and well-demonstrated aneurysmal dilatation (asterisk) at the origin of an aberrant right subclavian artery (ARSCA) as Kommerell diverticulum. (Arch: Aortic arch, LSCA: left subclavian artery, LCCA: left common carotid artery, RCCA: right common carotid artery)
Incidence and morphology of Kommerell diverticulum

KD vs. sex, age, and degenerative change
Patients were divided into two groups: patients with KD (O/M ratio ≥ 1.5) and those without KD (O/M ratio < 1.5). The sex distribution, age, O/M ratio and degenerative change were compared to see if significant differences existed. To analyze the prevalence of KD in different age groups, we further compared the KD incidence between those over 50 years old and those 50 years old and under.

ARSCA/ALSCA vs. sex, age, KD incidence, O/M ratio and degenerative change
We divided the patients into two groups, ARSCA and ALSCA, to compare the sex distribution, age, KD incidence, O/M ratio and degenerative change.

Age vs. aberrant subclavian artery diameters in the ostium and midline
The ASCA-O and ASCA-M were tested for correlation with age. A linear regression model was used to calculate the annual growth rates of ASCA-O and ASCA-M. The annual growth rates of ASCA-O and ASCA-M were further compared.

Intraobserver/interobserver agreement and analysis
To ensure this study was not biased by the measurement variation, we randomly selected 20 patients for intraobserver/interobserver agreement and variation analysis. The images of these 20 patients were measured twice by the same interpreting chest radiologist with a 1-week interval for intraobserver analysis. The repeat measurements included ASCA-O, ASCA-M and degenerative degree. Another independent chest radiologist with 8 years’ experience also measured the same parameters for interobserver analysis.

Statistical analysis
Statistical analysis was performed using commercially available software (SPSS 13.0 for Windows; SPSS, Chicago, IL, USA). Generally, a two-tailed test with $p < 0.05$ was considered
statistically significant. For expressing continuous variables, mean ± standard deviation was used. Data in the parentheses represent minimal and maximal values. For comparing the sex distribution and incidence of two groups, the Chi-square test was used. If the expected number was less than 5 in any cell, Fisher’s exact test was used instead. Since many articles [2, 3, 5] suggested the KD incidence seems to be higher for ALSCA patients than ARSCA patients, a one-tailed Chi-square or Fisher’s exact test was used in this part. For comparing continuous variables in the two groups, an independent t test was used. The Mann-Whitney U test was used for comparing ordinal variables such as degenerative change degrees. Pearson’s correlation was used for correlation analysis. A linear regression model was used to calculate the correlation coefficient, constant and $R^2$. The intra- and inter-observer agreements were also calculated by Pearson’s correlation. Intra- and inter-observer variations were defined as: (measurement 2 – measurement 1)*2/(measurement 2 + measurement 1)*100%. The agreement of ordinal variables was tested by kappa statistics.

RESULTS

Patient characteristics

During the five-year study period, after excluding the follow-up CT scans of the same patients, a total of 30,898 CT scans were performed on 30,898 patients. ASCA were found in 195 of them, indicating an incidence of 0.63% (195/30898). Four of them received only non-contrast scan due to history of contrast allergy. Thus, 191 patients were enrolled in the following analysis and 102 (53.4%) of them were female. The mean age was $28.0 \pm 26.6$ (0-91) years. ARSCA was found in 172 (90.1%) of them while ALSCA was found in 19 (9.9%) of them. The O/M ratio was $1.33 \pm 0.25$ (0.79-2.28). KD was found in 37 (19.4%) patients. The patient characteristics and degenerative changes are summarized in Table 1.

Table 1. Characteristics of patients with aberrant subclavian artery

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Data (n=191)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td>M: 89 (46.6%), F: 102 (53.4%)</td>
</tr>
<tr>
<td>Age (years)</td>
<td>28.0 $\pm$ 26.6 (0-91)</td>
</tr>
<tr>
<td>ARSCA : ALSCA</td>
<td>172 (90.1%) : 19 (9.9%)</td>
</tr>
<tr>
<td>Relationship with trachea and esophagus</td>
<td>All behind trachea and esophagus</td>
</tr>
<tr>
<td>ASCA ostium diameter (mm)</td>
<td>10.4 $\pm$ 4.6 (2.3-30.6)</td>
</tr>
<tr>
<td>O/M ratio</td>
<td>1.33 $\pm$ 0.25 (0.79-2.28)</td>
</tr>
<tr>
<td>Incidence of Kommerell diverticulum</td>
<td>37 / 191 (19.4%)</td>
</tr>
<tr>
<td>Degenerative change</td>
<td>Degree 1: 167 (87.4%)</td>
</tr>
<tr>
<td></td>
<td>Degree 2: 10 (5.2%)</td>
</tr>
<tr>
<td></td>
<td>Degree 3: 12 (6.3%)</td>
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<td></td>
<td>Degree 4: 2 (1.0%)</td>
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</tbody>
</table>

Table 2. Patient characteristics with and without Kommerell diverticulum

<table>
<thead>
<tr>
<th></th>
<th>With Kommerell diverticulum (n=37)</th>
<th>Without Kommerell diverticulum (n=154)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td>M: 21, F: 16</td>
<td>M: 68, F: 86</td>
<td>0.168</td>
</tr>
<tr>
<td>Age</td>
<td>$39.4 \pm 26.0$ (7-86)</td>
<td>$25.2 \pm 26.1$ (0-91)</td>
<td>$&lt;0.001^*$</td>
</tr>
<tr>
<td>O/M ratio</td>
<td>$1.73 \pm 0.21$ (1.51-2.28)</td>
<td>$1.23 \pm 0.14$ (0.79-1.48)</td>
<td>$&lt;0.001^*$</td>
</tr>
<tr>
<td>Degenerative change</td>
<td>Degree 1: 31</td>
<td>Degree 1: 136</td>
<td>0.427</td>
</tr>
<tr>
<td></td>
<td>Degree 2: 2</td>
<td>Degree 2: 8</td>
<td></td>
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<tr>
<td></td>
<td>Degree 3: 3</td>
<td>Degree 3: 9</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Degree 4: 1</td>
<td>Degree 4: 1</td>
<td></td>
</tr>
</tbody>
</table>

* $< 0.05$
found. The youngest patients with KD were several 7-year-old boys (n=5) and one girl (n=1).

**ARSCA/ALSCA vs. sex, age, incidence of KD, O/M ratio and degenerative change**

The sex distributions, O/M ratios and degenerative degrees of ARSCA and ALSCA showed no significant difference. However, a higher incidence of KD was found in ALSCA patients (36.8% vs. 17.4%, p = 0.049) (Table 3).

We conducted further analysis to determine if a specific subgroup contributed to the higher incidence of KD in ALSCA patients. We found that the difference in KD incidence was mostly attributable to patients older than 50 years. In this study, ALSCA patients older than 50 years had a 100% (4/4) incidence of KD formation (Table 4).

**Age vs. ASCA ostium and midline diameters**

The ASCA diameter at ostium (ASCA-O) was positively correlated with age (p < 0.001), as was the ASCA diameter at midline (ASCA-M) (p < 0.001). Using a linear regression model, the ASCA-O (mm) could be calculated by the formula 7.00 + 0.12*Age (R²=0.49), and the ASCA-M (mm) by the formula 5.53 + 0.08*Age (R²=0.56). The annual growth rate of the ASCA at ostium (0.12 mm/year) was 1.5 times that of the ASCA at midline (0.08 mm/year).

**Intraobserver agreement and variation**

For ASCA-O and ASCA-M measurements, the intraobserver agreements were good (R²=0.998 and 0.996, respectively, both p < 0.01) with the intraobserver variations both within 5.0% (-3.97 to 3.51%) and -4.59 to 4.69%). For degenerative change grades, the kappa value was 1.0, indicating total agreement.

**Interobserver agreement and variation**

For ASCA-O and ASCA-M measurements, the interobserver agreements were good (R²=0.993 and 0.995, respectively, both p < 0.01) with the interobserver variations all within 11.0% (-6.80 to 4.76%) and -7.27 to 10.85%). For degenerative change grades, the kappa value was 1.0, indicating total agreement.

**DISCUSSION**

Our study is the largest one to investigate the incidence and morphology of KD and ASCA in the current literature. We found several interesting results. First, the youngest KD patient in this study was 7 years old. Second, with increasing age, the diameters of the ASCA at ostium and the ASCA at midline both increased. And the growth rate of the ASCA at ostium was 1.5 times that of the ASCA at midline. Third, ALSCA patients had a higher incidence of KD formation as compared with ARSCA patients, especially patients older than 50 years. Fourth, all ASCA were found coursing behind the trachea and esophagus.

Our results have the following implications. First, KD, as previously believed [1, 2, 9, 10], is indeed a congenital disease since it is also prevalent in children with ASCA. However, with increasing age, the ASCA at ostium grows 1.5 times faster than the ASCA at midline, which means the aging process
would further enhance KD formation. Older ASCA patients do have a higher incidence of KD, and KD formation seems to be a mixed congenital and acquired process. Second, the risk factors for KD formation are ALSCA and age. In this study, ALSCA patients older than 50 years had a 100% incidence of KD formation. Third, the frequently quoted statement that 20% of ASCA can course in front of the trachea or between the trachea and the esophagus [13] should be considered to be wrong. In this study, all 191 ASCA were located behind the trachea and esophagus. Therefore, our study resolved these long-standing controversies.

It is controversial whether KD is a congenital [1, 2, 9, 10] or an acquired disease [11, 12]. Some consider KD formation as an atherosclerotic process [13]. Our study provides incidence evidence to resolve the controversy and confirms that KD formation is a combination of both the congenital and acquired processes. We further analyzed the atherosclerotic grades on CT and noticed that KD formation was not associated with degenerative processes such as mural calcification or thrombus. Thus, the acquired part of KD can’t be explained by the atherosclerotic process alone. The possible causes are hemodynamic shear force and pressure, and different elasticity due to different embryological origin.

In regard to the risk factors for KD formation, many clinicians found that KD seems to be more prevalent in patients with right aortic arch and ALSCA [3-5]. However, because of the limited case numbers in these case reports and case series, no statistical evidence can be gleaned. Because our study enrolled 191 patients with ASCA, the large sample size makes the observation statistically significant. KD is more prevalent in patients with ALSCA than ARSCA. It is possibly due to the traction force from the ligamentum arteriosus at the origin of the ALSCA [10] or due to its large volume of blood [17]. Furthermore, we found that the difference was mostly attributable to patients older than 50 years. In this study, if we combined the two risk factors, age older than 50 years and ALSCA, the incidence of KD was 100% (4/4).

Controversy also exists about the course distribution of ASCA. It has been repeatedly stated that 80% of ASCA course behind the esophagus, 15% between the trachea and esophagus and 5% in front of the trachea [13], but the original source of this statement is unknown. Several authors challenged the statement by citing their clinical experience with a limited number of cases and embryologic illegitimacy [6, 7, 13]. Our study is the first one to use a large sample incidence analysis to confirm that all ASCA course behind the trachea and esophagus. Therefore, the other previously accepted course distributions should be considered wrong. Any cases reported to have a different course should be treated as exceptional cases.

There are also limitations in this study. The retrospective study design has inherent selection bias. However, since this study was based on an 1,500-bed tertiary referral center, the result could still be applied to other general medical centers.

In summary, this is the largest study about the incidence and morphology of KD and ASCA, and we conclude that (1) all ASCA course behind the esophagus, (2) KD formation is a combination of congenital and acquired processes, and (3) ALSCA and age are the two risk factors for KD formation.

ACKNOWLEDGEMENT

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REFERENCES

Incidence and morphology of Kommerell diverticulum

Kommerell憩室：五年電腦斷層盛行率與型態研究

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本研究將分析異常鎖骨下動脈與 Kommerell憩室的盛行率與型態，以回答過去文獻中的爭議。

我們回溯閱讀近五年、30898位患者的胸腔電腦斷層，發現有195（0.63%）位患者有異常鎖骨下動脈。排除掉四個無注射顯影劑之檢查後，191位患者的影像被收入作進一步的分析。我們記錄患者的動脈分支狀況（「右主動脈弓並有異常左鎖骨下動脈」或「左主動脈弓並有異常右鎖骨下動脈」）、異常鎖骨下動脈的開口（ASCA-O）與位於中線處（ASCA-M）的直徑。ASCA-O/ASCA-M比值大於等於1.5被定義為 Kommerell憩室。我們並進一步分析以下參數之間的關係：年齡、性別、主動脈弓分支型態、ASCA-O、ASCA-M、O/M比值以及是否有 Kommerell憩室出現。

所有191位異常鎖骨下動脈患者，其異常動脈都是走在食道後方的。多數的 Kommerell憩室是在較為年長的患者身上發現。異常鎖骨下動脈之開口（ASCA-O）生長速率推估為0.12 mm/year，是中線處（ASCA-M: 0.08 mm/year）的1.5倍。異常左鎖骨下動脈的患者比起異常右鎖骨下動脈的患者有更高的 Kommerell憩室發生機會。50歲以上的異常左鎖骨下動脈患者有100%（4/4）的機會產生 Kommerell憩室。

本研究說明：(1) 所有異常鎖骨下動脈都走在食道後方、(2) Kommerell憩室的形成具有先天與後天混合的成分、(3) 異常左鎖骨下動脈以及年齡，是形成 Kommerell憩室的主要因素。