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Vertebral Tortuosity Index in Patients with Non-Connective Tissue Disorder-Related Aneurysm Disease

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WHAT THIS PAPER ADDS

Vertebral tortuosity index (VTI) has been described as a tool in patients with, or suspected to have, connective tissue disease, and has been shown to be associated with increasing severity of presentation. In this study, VTI was measured in patients with aneurysms who have no signs of connective tissue disease and the mean VTI was documented, as well as the disease patterns described. This may provide insight into the evolving understanding of the imaging phenotype of aneurysmal disease.

Objective: The vertebral tortuosity index (VTI) predicts increased risk of acute aortic events in patients with known genetic aortopathies. This study describes the VTI in a cohort of patients with non-connective tissue disorder-related large aneurysms.

Methods: Hospital imaging records from July 2012 to March 2016 were interrogated to identify patients with aneurysmal disease who had undergone computed tomographic angiography that included imaging of vertebral arteries. A control group of consecutive patients undergoing carotid and vertebral imaging was also assessed. VTI was calculated using the formula: [(centre-line distance) / (straight-line distance)-1] $\times$100 for all patients, and statistical analysis undertaken to determine whether measured VTI was statistically different in patients of younger age, with larger aneurysms, or an acute presentation. Comparison was made with patients who had no aneurysm disease.

Results: Sixty-five patients were identified with adequate imaging to assess the entire aorta, including vertebral arteries. The majority of patients were male (71%, 46/65) and mean age at the time of the CT scan was 71 years (SD 11.1 years). There were 11 patients under the age of 60 years in this cohort. The mean VTI was 33.17 (SD 20.43). There was no statistically significant difference between different territories of presentation (proximal vs. distal aneurysm, p=.94), age of patient (>60 years vs. <60 years, p=.2), or size of aneurysm (>6 cm vs. <6 cm, p=.09). Acuity of presentation was not predicted by a higher VTI (p=.69). The VTI in patients with aneurysms was higher than in patients without aneurysm disease (VTI = 16.1, p<.005)

Conclusions: An elevated VTI is consistently present in patients with degenerative aneurysms and has potential as a universally available predictive measurement. However, the increased VTI in the older cohort without connective tissue disease may not carry the same predictive value for acute presentations as has been demonstrated in younger patients with a known genetic basis for their aortopathy.

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INTRODUCTION

The understanding of aneurysm disease has evolved in the last two decades to incorporate a spectrum of phenotypes for genetic aortopathies that overlap.[1,2] Clinicians now observe arterial dilation and tortuosity occurring concurrently in patients with aneurysmal disease. Arterial tortuosity syndrome itself is associated with both lengthened arteries as well as aneurysms, and occurs after a mutation in SLC2A10,[3,4] leading to a loss of function in GLUT10, a glucose transporter which has been linked to alterations in the TGF-beta signalling pathway in some experimental models.[5] Arterial tortuosity also has been associated with Loeys Dietz syndrome, which is linked to alterations in TGF-beta signalling, and has been described in other connective tissue disorders as well.[6–8] Although the exact mechanism for arterial tortuosity is not fully elucidated, and different causes have been postulated to account for the decrease in the structural stability of the arterial wall,[9,10] it is becoming recognized as a common characteristic of non-syndromic aneurysms as well, and thus is of importance to the aortic surgeon.

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Arterial tortuosity can be used as a predictive variable when assessing the imaging phenotype of patients with aneurysmal disease. The SVS guidelines cite arterial tortuosity in the anatomic scoring system,[11] and it has been associated with adverse clinical outcomes following endovascular repair in the iliac territory.[12] Shirali et al. have found tortuosity in the aorta and the vertebral territories to be predictive of the risk of type B dissection.[13] Elevated vertebral tortuosity index has been shown to be associated with adverse cardiovascular events in patients with known connective tissue disorders.[14] Given the emerging importance of arterial tortuosity in patients with aneurysm disease, this study sought to describe the baseline vertebral tortuosity in patients with degenerative aneurysms and no known connective tissue disease.

METHODS

This study sought to review imaging on patients presenting to a single institution with findings of aneurysmal disease on computed tomography (CT) scan, including imaging of both vertebral arteries. This study did not fall under the auspices of the NHS Research Ethics committee according to Health Research Authority guidance; therefore it was registered with the Royal Free London NHS Foundation Trust as an audit, complying with local governance. Imaging databases were interrogated for patients with aneurysmal disease from July 2012 to May 2016. All patients with imaging of the entire aorta, that included the territory of vertebral arteries to base of skull were included. CT scans where the entire vertebral artery to the level of C2 was not visible were excluded from this analysis. Patients with known connective tissue disorder were excluded from the analysis.

A control group was collected from a list of consecutive patients who were referred for the study "CT angio aortic arch and carotid both" to the department of radiology at Royal Free London. The first 20 patients from this list of patients who met the criteria of having no known aneurysm disease, and who had two vertebral arteries visible for measurement were included in the control group.

A retrospective review was performed of the patients' in-hospital records and the electronic medical records. Patient characteristics including age, acuity of presentation, common comorbidities, presence of select medical treatments, and smoking status were collected when available. Where possible, contemporaneous information with the date of imaging was recorded.

CT scans were viewed using commercially available post processing software linked to the institutional PACS system for analysis and measurement (Aquarius Intuition, Terarecon, CA, USA). CT scans were loaded and an independent review of imaging data for each patient was performed by one investigator who underwent instruction in standardized methods for measurement of both aneurysm diameter, and vertebral tortuosity index (VTI). Validity of the measurements was assessed by blinded re-measurement of VTI by a senior investigator, and Pearson correlation coefficient was calculated to assess inter-observer agreement.

Aneurysmal disease was assessed using a standardized technique. Studies were reviewed in both multiplanar and in centre line of flow reconstructions. The maximum aneurysm diameter was measured in a plane perpendicular to the aneurysm at the level deemed to be greatest by the investigator. Aneurysm type was classified according to the Crawford classification system[15] based on the extent of anatomic disease rather than type of intended repair.

VTI was measured following the protocol described by Morris et al.[14] A centre line of flow spine was placed in both vertebral arteries for each patient from the base of the artery (either at the level of the subclavian or the level of the arch) to the C2 vertebral spine (Fig. 1). The abrupt change in direction at the level of C2 is predictable and thus the measurement was concluded proximal to this anatomic bend, or to the top of C2 where appropriate.[14] The actual length of the vessel was measured using the centre line of flow path, as calculated by proprietary algorithms in Terarecon software. Where necessary manual adjustments were made to account for the inability of the segmentation to differentiate artery from bone in some contrast densities. The linear distance between the base of the artery, and the level of the bend at C2 was also measured in the three-dimensional projection. The VTI was then calculated using the formula: [(centre-line distance) / (straight-line distance)-1] $\times$100 as previously described. To ensure the reproducibility of the technique, a blinded reviewer remeasured 20 random arteries to determine the Pearson correlation coefficient and assess the level of agreement. The highest VTI for each patient was used in the analysis.

Intended outcome analysis included assessment of mean VTI for patients with aneurysmal disease in different anatomic territories: proximal aneurysms were defined as any thoracic or thoracoabdominal aneurysms and distal

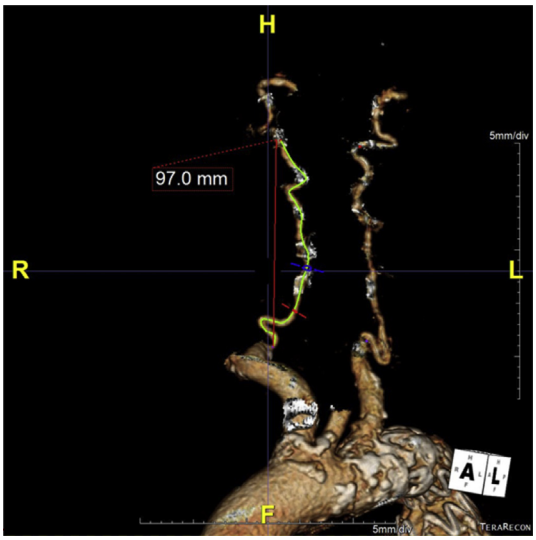

Figure 1. Straight line and tortuous line paths for determination of VTI in CT scans. The calculation is performed using the formula: [(centre-line distance) / (straight-line distance)$-$1] $\times$100. Given that the centre-line distance is 138 mm and the straight-line distance is 97 mm, the VTI is 42 in this patient.