Styloid and Hyoid Bone Proximity Is a Risk Factor for Cervical Carotid Artery Dissection

Dimitri Renard, MD; Souhayla Azakri, MD; Caroline Arquizan, MD; Bart Swinnen, MD; Pierre Labauge, MD, PhD; Vincent Thijs, MD, PhD

Background and Purpose—Carotid artery dissection (CAD) is more common with increased styloid process length. Our goal was to determine whether proximity of the styloid process and the hyoid bone to the internal carotid artery (ICA) was a risk factor for CAD.

Methods—We studied axial slices on computed tomography angiograms of 88 patients with nonaneurysmal CAD, from 88 age- and sex-matched controls without dissection, and from 32 nonage-/sex-matched nonaneurysmal vertebral artery dissection control patients. We measured the nearest distance between the ICA and both the styloid and the hyoid bones, blinded to clinical information and radiological reports.

Results—Styloid-ICA and hyoid-ICA distances were significantly shorter on the side of the CAD as compared with nondissection control patients ($P<0.0001$ for the styloid-ICA distance; and $P=0.0037$ for the hyoid-ICA distance). Styloid-ICA distances, regardless of the side of the dissection, were shorter in CAD patients compared with the nondissection control group (right side, $P=0.001$; left side, $P=0.0002$) and the vertebral artery dissection control group (right side, $P=0.0031$; left side, $P=0.0067$). Direct mechanical contact of the styloid with the ICA was more common in CAD patients.

Conclusions—Shorter distances between the styloid and ICA (and possibly also the hyoid and the ICA) are important risk factors for CAD. Further study is needed to determine whether dissections result from direct injury to the outer vessel wall of the carotid artery. **(*Stroke*. 2013;44:2475-2479.)**

Key Words: carotid arteries ■ dissection ■ hyoid bone ■ styloid

Carotid artery dissection (CAD) is an important cause of stroke in young adults. Major penetrating or nonpenetrating trauma to the artery is often lacking, but precipitating events involving hyperextension, rotation, or lateroversion of the neck are often reported by patients with CAD. These include whiplash injury, chiropractic manipulation, various sporting activities, neck movements, and severe coughing. The exact role of these precipitating events in the pathophysiology of CAD is unclear, but suggests a mechanical contribution.

The styloid process and hyoid bone, both in proximity to the internal carotid artery (ICA), have been implicated in the pathogenesis of CAD and nondissection carotid pathology in several case reports, including patients with Eagles syndrome.[1–10] The stylohyoid complex consists of the styloid process, the stylohyoid ligament, and hyoid bone. The anatomy of the styloid and the hyoid bone is variable. The stylohyoid complex syndrome refers to cervical and pharyngeal pain and discomfort associated with abnormalities of these structures. These abnormalities include elongated styloid processes or hyoid bones, ossifications of the stylohyoid ligament, or combinations of these pathologies. Symptoms are thought to result from irritation of the structures around this complex. Theoretically, the close proximity of the stylohyoid complex to the ICA may cause injury to the artery. Several hypotheses have been put forward, including the formation of dissection, direct compression resulting in stenosis or occlusion, or pressure-induced plaque formation or plaque rupture.[1–10]

Only 1 study has systematically investigated involvement of the styloid process in CAD.[11] In a case–control study of 38 patients with CAD, CAD was associated with a longer styloid process compared with age- and sex-matched controls. In addition, the styloid process was found to be significantly closer to the carotid artery contralateral to the CAD. The proximity of the styloid bone ipsilateral to the CAD was not studied because the authors considered measurement unreliable.

To our knowledge, the proximity of the hyoid bone to the ICA has never been systematically analyzed in patients with CAD.

If the styloid and the hyoid bone contribute to the pathogenesis of CAD through mechanical injury, the proximity

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(ie, the nearest distance between the bone structure and the carotid artery) may be a more important risk factor than the length itself of these bony structures. Longer styloid or hyoid bone structures might indirectly represent a closer anatomic proximity to the carotid arteries, but the distal end of these bony structures does not necessarily represent the area of closest contact to the artery.

Our aim was to analyze the distance between the ICA and both the styloid and the hyoid bones in a large group of patients with CAD, and controls.

Methods

Patient Selection

We performed a retrospective multicenter case–control study of CAD patients and age- and sex-matched controls without dissection and patients with vertebral artery dissection (VAD). Patients and controls were selected from stroke registries of 3 University Hospitals in Belgium and France between 2005 and 2012. Cases and controls had undergone computed tomography angiograms (CTA) as part of their routine workup of suspected cerebral ischemia or workup of local symptoms related to dissection (Horner syndrome, cranial nerve disease, or headache). Known iodine contrast allergy and renal insufficiency were contraindications for CTA in both the CAD and control populations. In all 3 centers, CTA was frequently used in both the initial workup (especially if MRI-MRA and duplex scanning did not lead to definitive diagnosis) and during follow-up. Ethics approval was obtained according to local regulations. Informed consent requirements were waived. Common cardiovascular risk factors were recorded in the CAD, VAD, and nondissection group. Potential risk factors for dissection (coughing, neck movement, minor trauma, etc) were not systematically recorded in the nondissection group and, therefore, not mentioned.

Cases

Cases were defined as patients with a unilateral ICA dissection. We excluded patients with radiological evidence of fibromuscular dysplasia and patients with pseudoaneurysmal forms of dissection. We also excluded patients with dissections of the common carotid artery (because styloid/hyoid bone proximity exists most often above the carotid bifurcation and, therefore, a mechanical role is less likely), patients with concomitant vertebral artery or contralateral ICA dissection, and patients with a recent history of penetrating trauma, neck surgery, and patients with aortic dissection.

CAD was confirmed in all patients by additional examinations (carotid duplex ultrasound, MR angiography, fat-saturated T1-weighted MRI, and rarely conventional angiography).

Controls

Two control groups were defined: (1) age- and sex-matched controls who underwent CTA and who did not have a history of carotid artery stenting or surgery and no CAD and (2) patients with a nonaneurysmal form of VAD. Age- and sex-matching were not performed in this last group. The VAD group was included because proximity of the bony structures to CAD might be just a bystander phenomenon common to all dissection patients. For anatomic reasons, it is unlikely that styloid and hyoid bone structures play a role in VAD.

Measurements

CTA was performed with different scans with slightly different parameters (Leuven-Belgium: Siemens Somatom Sensation 64, 120 kV, 95 mAS, 250 mm field of view, 512×512 matrix or Siemens Somatom Definition Flash 2011, 100 or 120 kV, 90 mAS, 250 mm field of view, 512×512 matrix; Nîmes-France: Siemens Somatom Definition AS+, 100 or 120 kV, 180 mAS, 500 mm field of view, 512×512 matrix; Montpellier-France: General Electric Lightspeed Vinyl Composition Tile 64, 120 kV, 250 mAS, field of view Head, 512×512 matrix), with slice thicknesses of the analyzed axial slices varying between 0.75 and 1.25 mm. CTA was done with the patient in supine and the head in neutral position. A single rater (D.R.) analyzed the nearest distance between the ICA and both the styloid and the hyoid bone on axial slices of CTA. We used the midpoint of the entire artery as a reference point (Figure 1), to avoid potential underestimation of the bone-ICA distance attributable to ICA dilatation resulting from CAD and because we found that even when the ICA was partially or completely occluded, this point could be reliably localized on CTA. When reliable visualization of the ICA was not possible because of severe stenosis or occlusion, subjects were excluded from analysis. Patients with a pseudoaneurysm were excluded from analysis because the distance between the carotid artery and the styloid/hyoid bone could not be assessed reliably.

We used the axial slice where the smallest distance was observed between the closest bone edge of the styloid or the hyoid bone and the midpoint of the entire artery. In cases where the carotid bifurcation was located above the level of the hyoid bone, the distance between the hyoid bone and the common instead of the ICA was measured. We obtained 4 distances for cases and controls (ie, right styloid-right ICA distance, left styloid-left ICA distance, right hyoid-right ICA distance, and left hyoid-left ICA distance).

We also recorded how frequently direct mechanical contact between the bony structure with deformation of the ICA was observed (Figure 2). If patients had undergone several CTA as part of their clinical follow-up, measurements were determined on the CTA that was performed most remotely in time from the initial clinical event. We recorded the time between symptom onset and the analyzed CTA.

Measurements were performed blinded to the clinical information and radiology reports. However, complete blinding was impossible because dissection-related radiological CTA abnormalities could be observed by the rater. To study consistency of the rater, 60 measurements were repeated after several weeks. The concordance coefficient, a measure of intrarater agreement proposed by Lin et al[12], was 0.92 (95% confidence interval, 0.88–0.96).

Statistics

We used ANOVA or the Student *t* test to detect statistical significant differences in distances between the styloid/hyoid bone and the ICA between the different groups, and Pearson's χ^2 test to detect significant differences between the occurrence

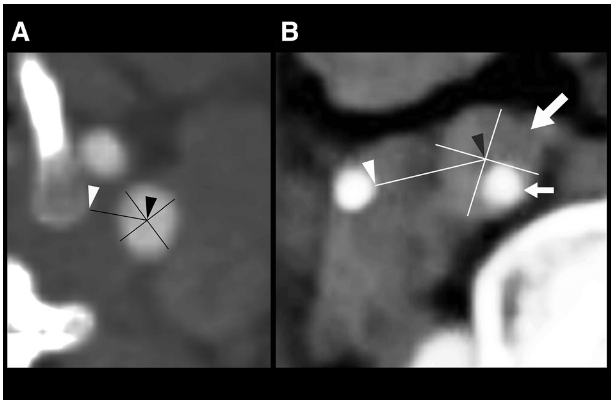

Figure 1. Axial computed tomography angiograms views showing the method used to calculate the distance in (**A**) a control patient between the closest part of the left side of the hyoid bone (white arrowhead) and the midpoint of the left internal carotid artery (ICA; black arrowhead), and in (**B**) a patient with right-sided carotid artery dissection between the right styloid bone (white arrowhead) and the midpoint of the partially occluded right ICA (black arrowhead). Inside the dissected ICA, the intramural hematoma (large arrow) and the circulating lumen (small arrow) can be seen.

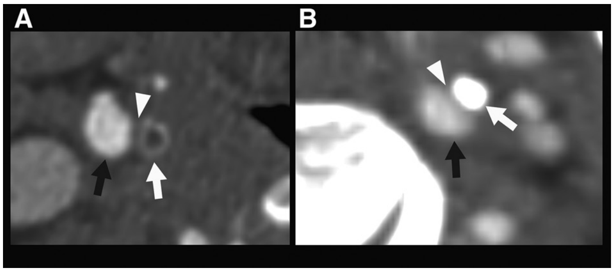

Figure 2. Axial computed tomography angiograms views showing 2 examples of suspected direct contact (arrowheads) between (**A**) the greater horn of the right side of the hyoid bone (white arrow) and the internal carotid artery (ICA; black arrow), and between (**B**) the left styloid bone (white arrow) and the left ICA (black arrow).

of direct mechanical effect of the styloid/hyoid bone structures to the ICA in the different groups. Two-sided *P* values of <0.05 were considered significant. Post hoc ANOVA comparisons were Bonferroni corrected.

Results

In the CAD group, 2 patients were excluded from analysis because of bilateral absent styloid bones, and 2 patients were excluded because reliable visualization of the carotid artery (because of occlusion) was not possible. In the nondissection control group, absent styloid bones were encountered in a patient, and reliable visualization of the carotid artery was not possible in another patient. These 2 patients were replaced with 2 other age- and sex-matched controls.

Patient characteristics in the patient and control populations are shown in the Table. The only significant differences were that patients with VAD were younger than those with CAD and controls, and that hypertension, hypercholesterolemia, and diabetes mellitus was more frequent in controls compared with CAD. The presence of these vascular risk factors was not associated with closer proximity to the styloid or hyoid (*P* values >0.10).

In the CAD group, the mean time between the first symptoms related to the CAD and the analyzed CTA was 174 days (SD, 326). We included and analyzed 88 CAD patients, 88 nondissection (age- and sex-matched with patients with CAD) control patients, and 32 (not age-/sex-matched) VAD control patients. The CAD group consisted of 58 men (66%) and 30 women (34%), with a mean age of 48 years (SD, 10.2). The control group had exactly the same number of men and women and a mean age of 49 years (SD, 9.7). The VAD group had 32 patients, 15 men and 17 women, with a mean age of 44 years (SD, 11.0). Figure 3 shows the comparison of the mean distances between the styloid/hyoid and the ICA in the different groups. Both styloid and hyoid bone structures were closer to the ICA in patients with CAD than in the nondissection control group (all differences were significant except for the left hyoid-left ICA distance) and the VAD control group (all differences were significant except for the right hyoid-right ICA), regardless on which side the CAD had occurred. There were no significant differences in styloid/hyoid-ICA distances between the nondissection control group and the VAD control group.

Figure 4 shows the distances of the styloid and hyoid at the side of the CAD and at the side opposite to the CAD in patients with CAD and in the control group. At the side of the CAD, both styloid and hyoid bone structures were significantly closer to the ICA compared with age-and sex-matched non-CAD controls. On the side opposite to the CAD, only the styloid-ICA distances were significantly smaller.

Within the CAD group, the styloid-ICA distances were significantly shorter (8.5 mm; 95% confidence interval, 2.2–14.8 mm) at the side of the dissection than at the nondissected side. There were no differences in hyoid-ICA distances (5 mm; 95% confidence interval, −5.0 to 15.1 mm).

A higher number of cases had direct mechanical contact of the styloid/hyoid bone structure with the ICA in the CAD group (n=20/88; 23%) compared with the nondissection control group (n=6/88; 7%; *P*=0.007) and the VAD control group (n=1/32; 2%; *P*=0.026). Of the 20 patients with CAD with bone contact, in 17/20 the side of the bone contact was concordant with the side of the CAD. In these 17 patients, 9 patients had no mural hematoma on the analyzed slice, 4 patients had mural hematoma producing stenosis, and 4 patients showed occlusion attributable to the mural hematoma.

Because positioning of the head may influence the distances between carotid and bony structures, we measured the angle between cerebral midline structures and the midline of C2 in the axial plane, as a measure of rotation between the skull and the cervical spine in a randomly selected subgroup of 20 patients with CAD and 20 controls. There was no difference between the rotation in the CAD group (mean angle, 3.2°; SD, 3.6) and the control group (mean angle, 3.7°; SD, 3.6; *P*=0.62).

Table. Patient Characteristics in the Patient and Control Populations

Variables	CAD	VAD	Control Group	*P* Value From ANOVA or χ^2 Test
Mean age (SD)	48 (10)	44 (11)	49 (10)	0.039*
Female sex, n (%)	30 (34)	17 (53)	31 (35)	0.138
Hypertension, n (%)	19 (22)†	9 (28)	41 (47)	0.002
Hypercholesterolemia, n (%)	18 (20)†	8 (25)	35 (40)	0.016
Diabetes mellitus, n (%)	0 (0)†	0 (0)	8 (10)	0.002
Active smokers, n (%)	23 (26)	9 (28)	29 (33)	0.604

CAD indicates carotid artery dissection; and VAD, vertebral artery dissection.

*VAD group is significantly younger than control group.

†Hypertension, hypercholesterolemia, and diabetes mellitus are significantly more frequent in control group compared with CAD. No significant differences between VAD and control groups in terms of risk factors.

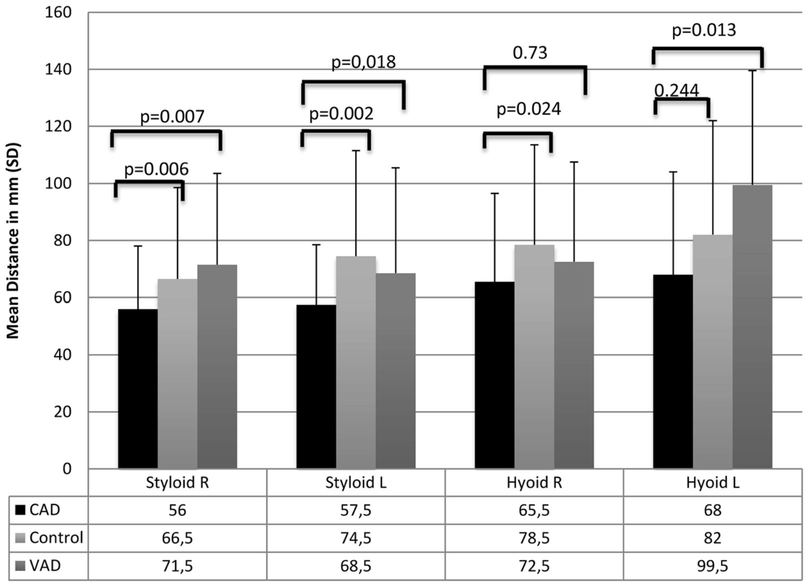

Figure 3. Comparison of the mean distances between the styloid/hyoid and the internal carotid artery in the 3 different groups. All ANOVA comparisons were significant. There were no significant differences between the vertebral artery dissection (VAD) groups and controls. Post hoc Bonferroni–corrected *P* values are shown. CAD indicates carotid artery dissection.

Discussion

Our study lends support to the hypothesis that anatomic factors predispose to the occurrence of CAD. Our results are in line with a previous study, which showed smaller styloid-ICA distances contralateral to the CAD side.[11] In that study, direct measurement of the distance between the bone structure and the affected vessel was not performed because the authors considered this unreliable. We confirmed the finding of a shorter distance opposite to the site of the dissection in a larger study population, but also showed that the styloid and hyoid bone were closer to the dissected artery. Our findings were specific to patients with CAD and not a general feature of patients with dissection because distances in patients with VAD did not differ from controls. The fact that both the previous study and our study point to the same conclusions despite the use of a different methodology reinforces the idea of a potential true mechanical role of the styloid process in, at least some, patients with CAD.

We consider our measurement method to be reliable. In the majority of our patients with CAD, the analyzed CTA was performed late after the acute phase of CAD. Therefore, intramural hematoma, stenosis, and occlusion were unfrequent, and reliable measurement of the styloid-ICA and the hyoid-ICA distances could be performed. We moreover excluded patients with pseudoaneurysmal forms of CAD or fibromuscular dysplasia in which measurements would be more difficult. We did not measure the distance to the outer wall of the CAD but instead the distance to the midpoint of the carotid artery, which is less likely to be displaced. The intrarater consistency was very high. Our findings, however, may have been influenced by the few patients with associated mural hematoma, which may alter carotid-styloid/hyoid distances. Further longitudinal study is needed to determine whether distances between the carotid and styloid/hyoid evolve over time.

If anatomic factors contribute so strongly to CAD risk, one could wonder why CAD recurrence is so rare.[13–15] This could be partly attributable to underestimation of the true rate of (asymptomatic) CAD recurrences because studies with long-term imaging follow-up are scarce. Because cervical artery dissection seems to be associated with a multitude of potential risk factors (including underlying connective tissue disorder, infection, migraine, hyperhomocysteinemia, genetic factors, and other environmental factors), styloid/hyoid proximity may be just one of several environmental factors playing a role in CAD. Potentially, only the temporal co-occurrence of certain head positions, movements, and head or cervical trauma in patients with pre-existing proximate styloid/hyoid bones create the unfavorable mechanical conditions leading to direct ICA injury provoked by the styloid/hyoid.

Our study has some limitations: we did not include a consecutive series of patients with dissections because CTA was not performed systematically in the 3 participating centers. Complete blinded measurements are impossible and might

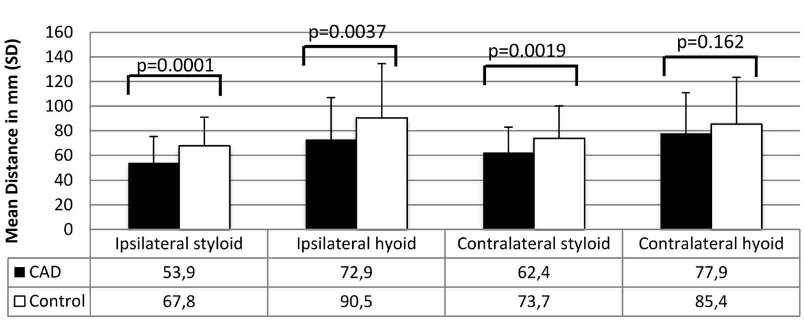

Figure 4. Distances of the styloid and hyoid at the side of the carotid artery dissection (CAD) and at the side opposite to the CAD in patients with CAD and in the control group.

have influenced the rater. Therefore, our findings need to be replicated in other cohorts.

Our study implies that the anatomic relationship between the stylohyoid complex in relation to the carotid artery should be examined more carefully in patients with CAD. Whether there is a role for surgery on the styloid or hyoid bone in management of CAD requires further study and cannot be recommended at present.

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Disclosures

None.

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Table.

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*VAD group is significantly younger than control group
** Hypertension, hypercholesterolemia and diabetes are significantly more frequent in control group compared to CAD. No significant differences between VAD and control groups in terms of risk factors.