

Transient Perivascular Inflammation of the Carotid Artery (TIPIC): Vascular Ultrasonography Role

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Transient Perivascular Inflammation of the Carotid Artery (TIPIC syndrome) is a rare and underdiagnosed condition. In this article, we describe a case of TIPIC syndrome, including topics such as clinical evaluation, indication and findings of vascular ultrasonography (VUS), treatment and follow-up, with a brief review of the literature.

Case Report

Male patient, 53 years old, complaining of progressive unilateral neck pain, which appeared spontaneously, in association with edema, local redness, and great sensitivity to palpation. The patient's past history included radiotherapy for about 5 years in the neck and face region, on the opposite side of the current complaint. The patient was a non-smoker and had no other significant risk factors for atherosclerotic disease. One year before the current clinical picture, the patient had undergone VUS of the carotid arteries, in a routine evaluation for follow-up after radiotherapy, the result was within normal limits.

Given the clinical picture, a new VUS examination was requested, which included the standard protocol for evaluating the extracranial common, external, and internal carotid arteries (ACC, ICA and ACE). Special attention was given to extracarotid (perivascular) tissues due to the history of radiotherapy. The B-mode US showed extensive hypoechoic and homogeneous involvement of the tissues, including the arterial wall at the level of the carotid bifurcation, expanding to the proximal ICA (Figure 1A). The peak systolic velocity and end-diastolic velocity (PSV/EDV) were high: PSV/EDV = 288/129 cm/s (Figure 1B). Local stenosis measured in the transverse plane of the

image was estimated at 74% (Figure 1C), consistent with the Doppler velocity data. The extent of the affected area was 5.57 mm compared to a lumen of 1.96 mm. Magnetic resonance imaging (MRI) and angioresonance (MRA) showed irregular thickening, with contrast enhancement, in the proximal ICA and bifurcation, corroborating the VUS findings (Figures 2A and 2B).

The patient underwent drug treatment with corticosteroids for 2 weeks with significant improvement in symptoms.

The VUS of the carotid arteries was repeated after 4 months, this second exam being essential for the diagnostic conclusion. Ultrasound examination showed that, after treatment, there was a reduction in the degree of stenosis to approximately 50% (Figures 3A and 3B). The ICA luminal diameter increased from 1.96 mm to 3.19 mm. PSV decreased from 288 cm/s to 132 cm/s and EDV decreased from 129 cm/s to 62 cm/s (Figure 3C).

Due to clinical improvement and VUS data after treatment, the diagnosis of TIPIC syndrome was considered.

Discussion

TIPIC syndrome is a rare and underdiagnosed disease, supposedly of an inflammatory nature.¹ Its etiology and pathophysiology are unknown, and it may be part of an autoimmune process that has not yet been clarified. Prevalence was estimated at 2.8%, in individuals with acute onset neck pain, with a slight male predominance^(1,5,11) and a mean age of 48 years. Its evolution suggests that it is benign, with resolution of the symptoms in approximately 2 weeks, either spontaneously or with the use of drugs such as non-hormonal anti-inflammatory drugs, corticosteroids and acetylsalicylic acid.¹⁻³

Laboratory tests can be normal or nonspecific, and the diagnosis is made by clinical presentation and imaging tests.¹ VUS is capable of identifying vascular and perivascular alterations in TIPIC syndrome, in which one of the characteristic alterations is the diffuse and homogeneous increase in the echogenicity of the perivascular fatty tissue at the site of pain, causing attenuation of the sound beam, as well as the presence of a self-limited intimal soft plaque could be related to the carotid inflammatory process.^{1,2} Other imaging methods that can also be used for the

Keywords

TIPIC; Carotid artery; arteritis

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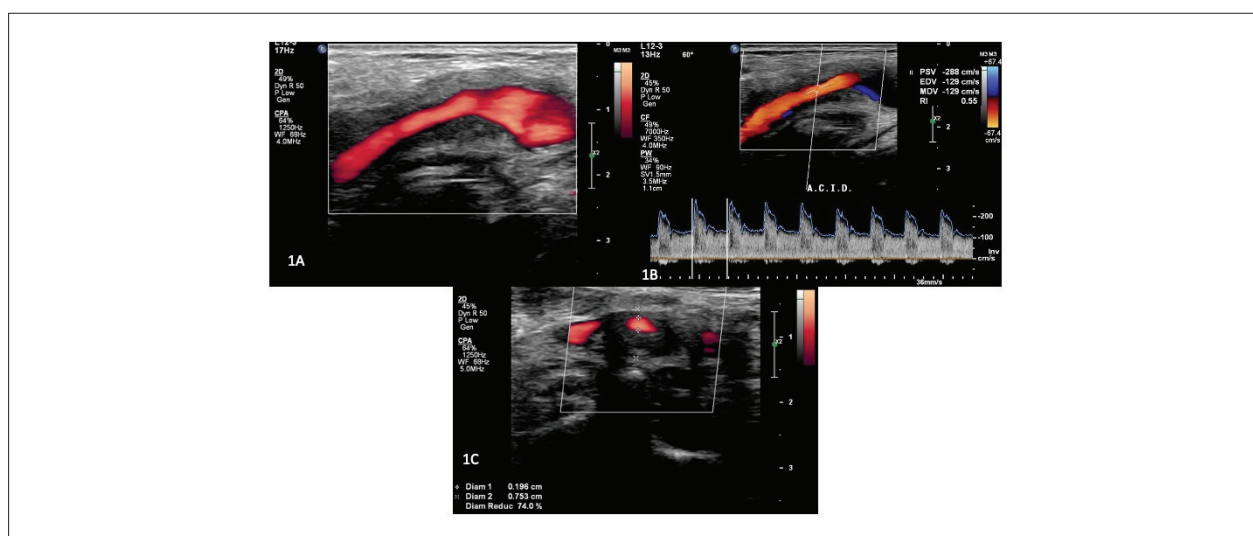


Figure 1 – Transient Perivascular Inflammation of the Carotid Artery (TIPIC syndrome) is a rare and underdiagnosed condition. In this article, we describe a case of TIPIC syndrome, including topics such as clinical evaluation, indication and findings of vascular ultrasonography (VUS), treatment and follow-up, with a brief review of the literature.

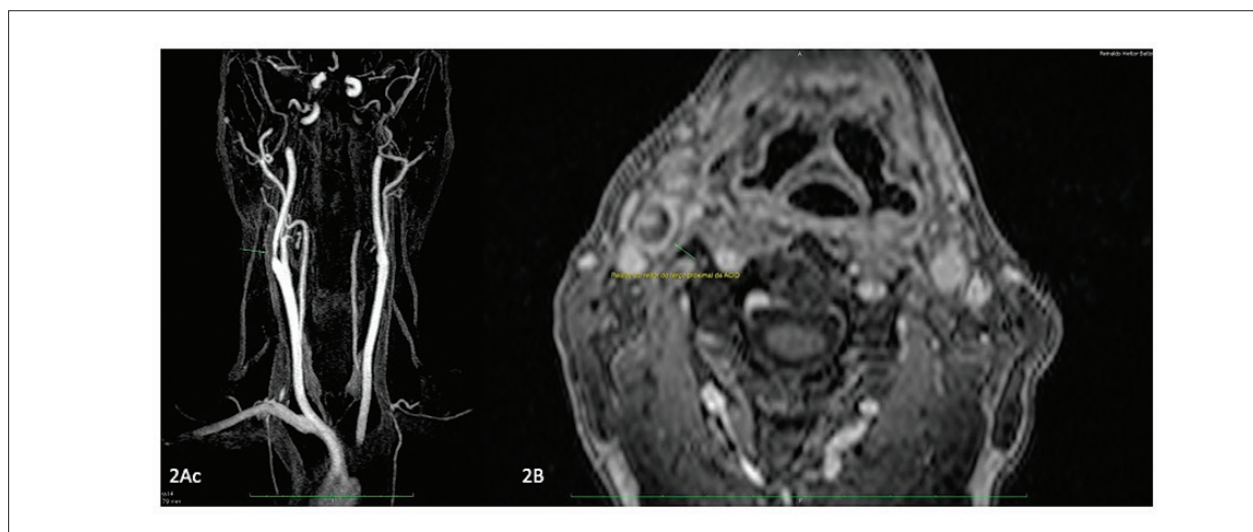


Figure 2 – MRI and magnetic resonance angiography (MRA). A) MRA showing a 70% reduction in the lumen of the proximal ICA; B) Irregular thickening with contrast enhancement in the periphery of the ICA. Cortesy: Fernando Santos Emerick Gomes M.D. Federal University of Espírito Santo.

diagnosis of TIPIC and that contribute to the sonographic findings are computed tomography (CT), MRI and NMR.²⁻⁴

A retrospective multinational observational study, involving 72 patients, discussed findings such as thickness and longitudinal extent of the disease in ultrasound images, with the purpose of evaluating the clinical behavior and characteristics at VUS in the TIPIC syndrome.² The authors reaffirmed the use of the previously proposed term TIPIC, replacing the term “carotidynia” for cases such as the one described in this article.^{1,2}

Lecler et al.¹ proposed 4 major diagnostic criteria for TIPIC: 1) presence of acute pain along the course of the carotid artery, which may radiate to the head; 2)

eccentric perivascular infiltration on imaging; 3) exclusion of another diagnosis, whether vascular or non-vascular; 4) improvement in 14 days with anti-inflammatory treatment or spontaneous. And yet a minor criterion: presence of self-limited intimal soft plaque.

Potential differential diagnoses include acute carotid artery dissection, Takayasu arteritis, giant cell arteritis, other vasculitis (including radiation-induced arteritis), jugular vein thrombosis, lymphadenitis, submandibular gland disorder, and tumors.^{1,2}

Lately, two case reports were published that presented similar clinical characteristics, however, one of them was affected by COVID-19. Both were male individuals, 38

Case Report

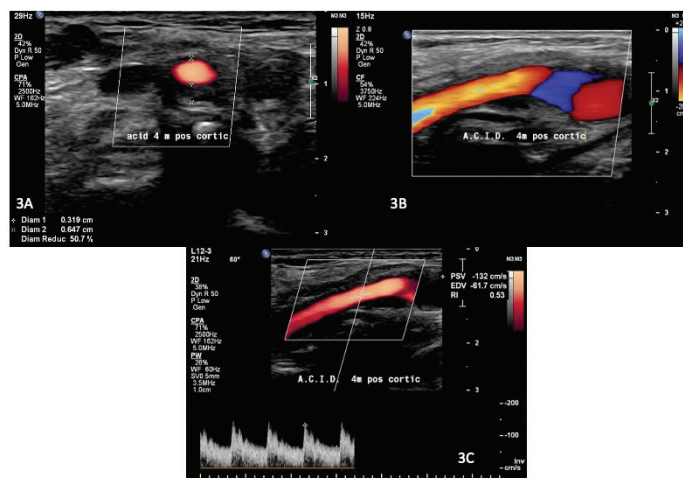


Figure 3 – VUS findings of the TIPIIC case, 4 months after diagnosis and treatment. A) Cross-sectional image in B-mode and power Doppler, with measurement of the degree of stenosis in the proximal ICA; B) Thickening reduction covering the wall of the proximal ICA, from the carotid bifurcation, with color Doppler flow; C) Pulsed Doppler of the proximal ICA demonstrating a reduction in flow velocity values.

and 45 years old, with thickening of the arterial wall of the ACC and carotid bifurcation on VUS. B-mode VUS imaging demonstrated hypoechogenic homogeneous tissue, without significant arterial stenosis, and the findings were corroborated by CT and magnetic resonance.^{3,4} As in our case, local signs and symptoms in the neck region improved within two weeks after treatment.

The VUS performed 4 months after the beginning of the condition in our patient showed a substantial improvement in the alterations, compared with the alterations detected in the diagnostic ultrasound examination. Such findings are consistent with the data described in the literature, and there may be complete remission of the ultrasonographic alterations, although there may be residual lesions.¹⁻³ One explanation, according to the histopathological study, would be the early development of fibrosis associated with chronic and low-grade inflammatory activity.¹

Ultrasound Tissue Characterization Analysis can use tissue brightness to identify cases of TIPIIC.⁵ The technique used by Lal et al.⁵ stratifies the B-mode image using reference points between 0-190 in differentiating blood -adventitia. Adventitia may not be clear in TIPIIC cases, but tissue characterization can separate homogeneous TIPIIC findings from heterogeneous findings suggestive of atherosclerotic plaque. There is also the prospect of using contrast-enhanced ultrasound (CEUS) to investigate TIPIIC findings, just as CEUS has been promising for the detection of flow on Doppler arising from neovascularization within carotid plaques^{2,6} and also in cases of carotid membranes.⁷

Conclusion

VUS, clinical history and physical examination documented a rare case of TIPIIC, which was confirmed by MRI. Treatment results were consistent with literature data. The etiology of the TIPIIC syndrome still requires further investigation for its better elucidation.

Author Contributions

Conception and design of the research: Barros FS, dos Santos SN, Cunha SS; Acquisition of data and analysis and interpretation of the data: Barros FS; Writing of the manuscript and critical revision of the manuscript for intellectual content: Barros FS, dos Santos SN, Barros DS, Cunha SS, Albriker ACL, Petisco ACP.

Potential Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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Study Association

This study is not associated with any thesis or dissertation work.

Ethics Approval and Consent to Participate

This article does not contain any studies with human participants or animals performed by any of the authors.

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