Transient Perivascular Inflammation of the Carotid Artery (TIPIC) Syndrome: Does Ultrasound Decipher the Myth?

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Abstract
Transient Perivascular Inflammation of the Carotid Artery (TIPIC) syndrome; is a rare, under-diagnosed clinico-radiologic entity comprising of acute unilateral neck pain and tenderness over the affected carotid bifurcation. In patients with TIPIC syndrome or idiopathic carotidynia, diagnostic imaging demonstrates a characteristic transient perivascular soft tissue thickening at the level of the affected carotid bifurcation. This article presents a short review of literature and highlights the role of Ultrasound in the diagnosis and follow-up of patients with a presumptive diagnosis of TIPIC syndrome.

Keywords: Neck pain; Syndrome; Carotid artery diseases; Diagnosis; Ultrasound

Abbreviations: TIPIC- Transient Perivascular Inflammation of the Carotid Artery; IC- Idiopathic carotidynia; IHS-International Headache Society; US- Ultrasound; MRI- Magnetic Resonance Imaging; CT- Computed Tomography; CTA- Computed Tomography angiography

1. Transient Perivascular Inflammation of the Carotid Artery (TIPIC)
Transient Perivascular Inflammation of the Carotid Artery (TIPIC), a newly classified syndrome of neck pain and tenderness described by Lecler et al., is diagnosed on the basis of four major criteria and one minor criterion (Table 1) [1].
### Table 1: Diagnostic criteria proposed by Lecler et al. for the diagnosis of TIPIC syndrome.

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<th>Major criteria</th>
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<td>1. Presence of acute pain overlying the carotid artery, which may or may not radiate to the head</td>
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<td>2. Eccentric perivascular infiltration (PVI) on imaging</td>
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<td>3. Exclusion of another vascular or nonvascular diagnosis with imaging</td>
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<td>4. Improvement within 14 days either spontaneously or with anti-inflammatory treatment</td>
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<table>
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<th>Minor criterion</th>
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<td>1. Presence of a self-limited intimal soft plaque</td>
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TIPIC was previously referred to as carotidynia and was first described by Fay in 1927 [2]. It was classified as idiopathic carotidynia (IC) by the International Headache Society (IHS) Classification Committee in 1988 [3]. However, in a review published in 1994, Biousse and Bousser dismissed carotidynia to be a myth and criticized the paucity of diagnostic imaging findings corroborating with the clinical presentation in suspected cases [4]. IC was thereafter removed from the IHS classification in 2004.

On the basis of case reports published over the recent years, TIPIC is being again recognized as a distinct clinico-radiologic entity. It is associated with characteristic imaging features of transient perivascular soft tissue inflammation at the level of the affected carotid bifurcation [1, 5-7]. Even though chronic inflammation of the affected carotid arterial adventitia has been implicated as the cause for TIPIC/IC; laboratory markers of inflammation are often normal [1, 5-7].

TIPIC usually presents as unilateral disease. However, Young et al. have described a unique case of bilateral metachronous disease in their article published in Clinical Imaging [8]. Takamura et al. have reported a rare recurrent case of TIPIC syndrome with a temporary carotid plaque in a young male [6]. I have encountered and reported a case of recurrent unilateral TIPIC syndrome with a past history of transient symptoms on the contralateral side of the neck [9]. Imaging techniques such as ultrasound (US), magnetic resonance imaging (MRI), computed tomography (CT) imaging, CT angiography (CTA), and [18F] fluoro-deoxy-glucose positron-emission tomography-CT have been employed for establishing the diagnosis of this benign, self-limiting condition [1,10].

In 2005, Arning reported a consistently similar ultrasound pattern of hypoechoic perivascular soft tissue thickening along the carotid bulb, exactly in the region of tenderness along with slight narrowing of the affected vessel lumen; in six patients suspected of carotidynia [11]. Regression of the pericarotid thickening was seen on follow-up US imaging in these patients. Similar findings were confirmed by Kuhn et al. in the research article published in...
Cephalalgia in 2006 [12]. More recently, Abrahamy et al. and Takamura et al. have demonstrated in their respective case studies that in a clinical setting of TIPIC syndrome, ultrasound with Doppler analysis serves as a first line imaging investigation [5, 6]. US shows the characteristic peri-carotid soft tissue thickening along with normal vascular hemodynamics.

The largest multicenter series of patients suspected to have carotidynia was investigated by Lecler et al. in 2017 [1]. In this case series, the authors demonstrated the characteristic peri-vascular imaging features on various imaging techniques including US, CT, CTA and MRI. The investigators also reported the presence of an associated intimal plaque in a good number of these patients (58%) and enlisted this as a minor criterion for diagnosis of TIPIC syndrome. On follow-up imaging, the plaque had resolved in some of the patients. Few other case reports by Coulier et al. and Takamura et al. have also documented a self-resolving intimal plaque along the affected carotid artery [6, 7]. However, it has been an inconsistent finding across the literature. The role of US in follow-up of patients with TIPIC syndrome has been documented by the above mentioned authors [1, 6, 7, 9].

In the case reported by Coulier B, contrast-enhanced ultrasound (CEUS) showed a normal capillary distribution of micro bubbles in the hypoechoic peri-vascular thickening [7]. A recent retrospective analysis of ultrasound images of five patients with TIPIC syndrome, performed by Rafailidis et al. revealed contrast enhancement of the perivascular lesions on CEUS, similar to the enhancement noted on contrast enhanced CT and MRI [13]. Lecler et al. and Coulier et al. observed that apart from confirming the US imaging findings; MRI plays a crucial role in diagnosing other vascular conditions mimicking carotidynia, such as giant cell arteritis, thrombosis, fibromuscular dysplasia, carotid dissection, aneurysm and lymphadenitis [1, 7]. Patients with TIPIC syndrome respond to anti-inflammatory medication [5-9]. Prompt and accurate diagnosis of this benign condition, thus facilitates appropriate treatment of the suspected cases and also avoids unnecessary investigations. It is therefore worthwhile to keep TIPIC syndrome in mind when examining patients complaining of unilateral neck or facial pain.

In the current clinical practice, with the introduction of the recently proposed diagnostic criteria; correlation of clinical presentation, laboratory findings and imaging features clinches the diagnosis of TIPIC syndrome. Ultrasound is a non-invasive, cost-efficient, widely available imaging technique that plays a pivotal role in the preliminary imaging diagnosis as well as follow-up of TIPIC syndrome.

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Conflict of Interest
None

References

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