

CASE REPORT

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Multimodality imaging of transient perivascular inflammation of carotid artery (TIPIC) syndrome: a case report

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Abstract

Background Transient Perivascular Inflammation of Carotid artery syndrome is a rare clinicroadiological entity characterized by inflammation of the carotid artery wall with surrounding perivascular inflammatory changes. This is a self-limiting condition and necessitates awareness and high degree of suspicion to differentiate from other serious pathologies involving the carotid artery.

Case presentation A middle aged asian male patient presented with acute onset of intense pain in the right lateral aspect of neck. Ultrasonography and Magnetic Resonance Imaging showed wall thickening of the common carotid artery and carotid bulb with surrounding inflammation in the perivascular soft tissues. He was treated with brief course of oral anti-inflammatory medications. The patient's symptoms improved and imaging findings resolved in 2 weeks.

Conclusions Being infrequently encountered in clinical practice, recognition of transient perivascular inflammation of carotid artery syndrome by the physician is of prime importance. Prompt radiological investigation and follow-up imaging are crucial for accurate diagnosis, thereby leading to appropriate treatment.

Keywords TIPIC, Carotid artery, MRI, Ultrasonography, Case report, Carotidynia

Background

Transient perivascular inflammation of carotid artery (TIPIC) syndrome is a rare self-limiting inflammatory condition which presents with acute pain in the side of the neck. The term TIPIC syndrome was coined by Lecler et al. [1]. The condition was previously referred to as carotidynia or Fay syndrome (named after Dr. Fay), which is a pain syndrome involving the carotid artery exacerbated by palpation. In 2004, the term 'carotidynia' was

removed from the International Classification of Headache Disorders [2] due to lack of specificity.

The etiology and pathogenesis of TIPIC syndrome is still unclear; however it appears to be inflammatory process involving the carotid wall including the adventitia. Histological examination done in limited studies revealed predominantly lymphocytic proliferation with scattered neutrophils and early fibrosis in the vessel wall [3].

TIPIC syndrome commonly presents a unilateral neck pain. The patient usually presents with acute onset of throbbing pain and swelling in the side of neck adjacent to the thyroid cartilage. Other uncommon symptoms are referred pain in head and neck region, vertigo and other transient neurological manifestations [4]. The inflammatory markers like C-reactive protein & erythrocyte sedimentation rate may be elevated but are nonspecific.

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Case presentation

A 37-year-old Asian male patient came to the hospital with complaint of right sided neck pain for 3 days, throbbing in nature and non-radiating. No personal history of diabetes, hypertension, connective tissue disorders, any recent viral illness or similar episodes in the past.

Clinical examination showed mild swelling and tenderness in the right side of the neck adjacent to the thyroid cartilage. Laboratory investigations revealed slightly raised C-reactive protein (15 mg/L) and normal erythrocyte sedimentation rate (10 mm/h). Complete blood count and differential leucocyte count were within normal limits. He was referred to Radiology department for USG of neck and followed by MRI.

In USG (Fig 1A), the distal CCA and carotid bulb shows hypoechoic wall thickening at the exact location of pain. The thickening was seen predominantly involving the anterolateral aspect of the wall for about 50% of the circumference. The maximum thickness of the wall measured 8 mm and involved a segment of 2.5 cm of distal common carotid artery and carotid bulb. There was no evidence of any luminal narrowing, intimal plaques or calcification in the CCA. On color Doppler (Fig 1B), normal color filling and spectral waveform were observed.

In MRI (Fig. 2A and B), wall thickening of distal CCA and carotid bulb was demonstrated and it appeared hypointense on T1 weighted sequence and hyperintense on T2 weighted fat suppressed sequences. The thickening

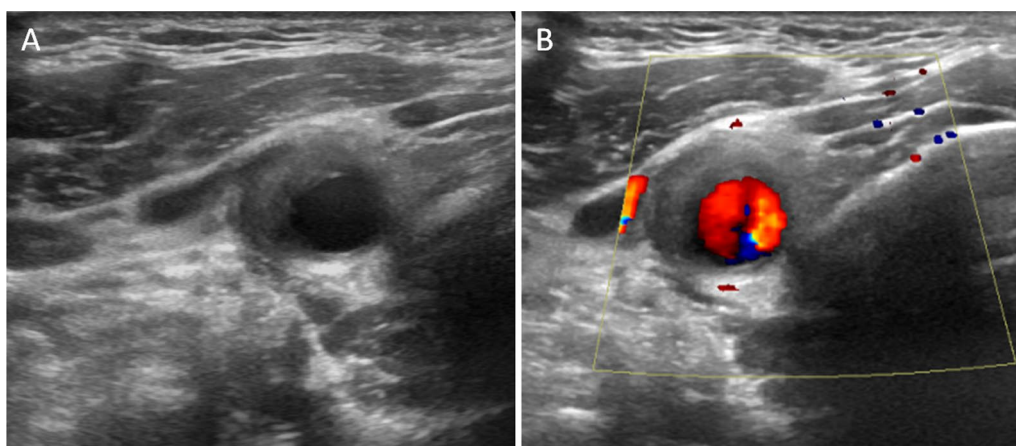


Fig. 1 A and B: Gray scale ultrasonography (A) shows eccentric thickening of the lateral wall of distal CCA with adjacent inflammatory changes and anechoic lumen. Doppler study (B) shows normal color filling of the lumen

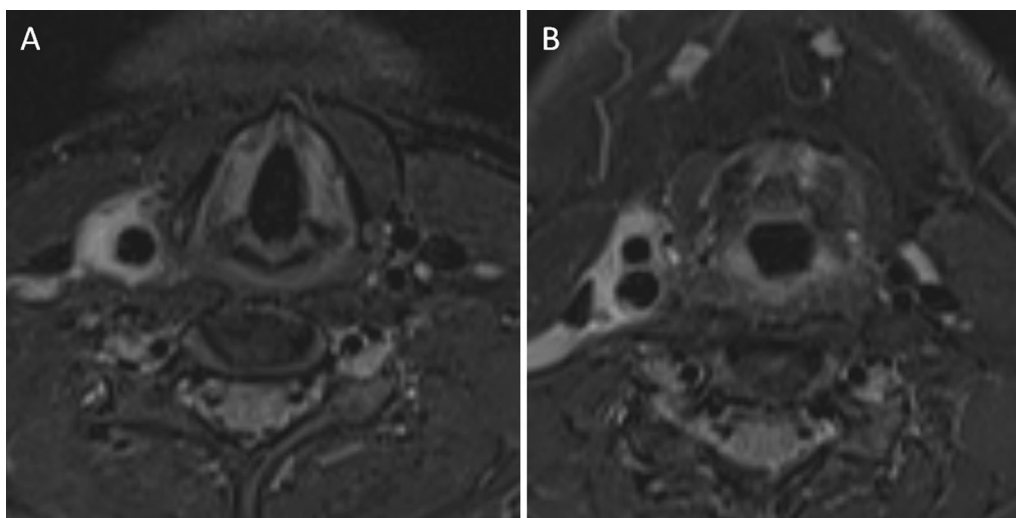


Fig. 2 A and B: MRI T2W fat suppressed images show hyperintense wall thickening of the distal right CCA (A) and carotid bulb (B) without luminal narrowing. Inflammation is also seen in the perivascular soft tissues. Note made of normal CCA and carotid bulb on the left side

was seen involving about half of circumference of distal CCA and carotid bulb in the anterolateral wall. Normal flow void was present in the lumen without any stenosis. T1 weighted fat suppressed post-contrast images (Fig. 3) showed moderate homogenous enhancement of the wall thickening and surrounding perivascular soft tissues. Few small discrete ovoid subcentimetric lymph nodes were noted in bilateral level II, III cervical groups. The contralateral CCA appeared normal.

Based on the imaging, diagnosis of TIPIC syndrome was made considering acute onset of symptoms and typical findings. The main differential diagnosis considered was vasculitis like Takayasu arteritis. Subsequent to the radiological diagnosis of TIPIC syndrome, the patient was explained about the condition and reassured of its transient nature. He was put on a short course of oral tablet Etoricoxib 60 mg once a day for 10 days and advised follow-up after 2 weeks. The patient's symptoms improved dramatically in a week and follow-up USG done at 2 weeks revealed complete resolution of the CCA wall thickening.

Discussion

Transient perivascular inflammation of carotid artery (TIPIC) syndrome is an idiopathic inflammatory disorder characterized by inflammation of the wall of carotid artery and surrounding soft tissues. Wall thickening is frequently seen involving the distal CCA and carotid bulb.

USG is the initial imaging modality of choice in diagnosing TIPIC syndrome which shows wall thickening involving the carotid artery typically located at

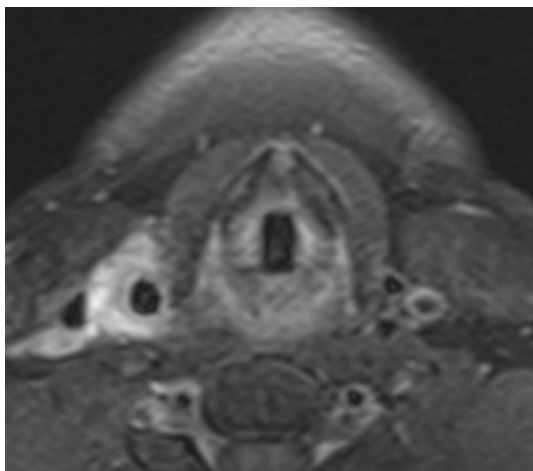


Fig. 3 MRI T1W fat suppressed post-contrast image shows enhancing wall thickening of the distal right CCA and inflammation in the surrounding perivascular soft tissues. Note made of normal left CCA

the bifurcation with associated probe tenderness and perivascular changes. Perivascular infiltration is seen as soft amorphous tissue replacing the fat surrounding the carotid artery. Vascularization of perivascular inflammation can be assessed by Doppler imaging [1]. Majority of the cases show minimal or no luminal narrowing and normal color filling in the lumen. Small reactive cervical lymph nodes may be present adjacent to the lesion. CT & MRI also demonstrate inflammation in the carotid wall and in the perivascular soft tissues. FDG PET done in few patients show increased uptake in the thickened wall and perivascular soft tissues.

Lecler et al. proposed 4 major criteria [1] to diagnose TIPIC syndrome which are presence of acute pain overlying the carotid artery, eccentric perivascular infiltration on imaging, exclusion of another vascular or nonvascular diagnosis with imaging and improvement within 14 days either spontaneously or with anti-inflammatory treatment. Additionally, a minor criterion could be the presence of a self-limited intimal soft plaque. Our case fulfilled all the major criteria mentioned above.

Though TIPIC syndrome is a distinct clinicoradiological entity, it is often a diagnosis of exclusion. Neck pain may be caused by multiple vascular and non-vascular etiologies. The differential diagnosis for a vascular cause includes large vessel vasculitis such as Takayasu arteritis, other autoimmune disorders, thrombosed aneurysm or dissection [5], hence a thorough personal history, general assessment and baseline investigations are essential.

Management is conservative with a short course of nonsteroidal anti-inflammatory drugs or steroids to accelerate the recovery. Spontaneous regression of the symptoms and imaging findings have also been reported. A follow-up USG is advised at the end of 2 weeks which usually shows complete resolution of the lesion. Some studies show delayed resolution of inflammatory changes and need for further follow-up imaging [6]. Being a novel entity with limited knowledge about etiopathogenesis and literature data, long-term follow-up is necessary to look for any association with development of plaques or any other long-term sequelae.

Conclusions

TIPIC syndrome is a rare self-limiting inflammatory condition involving the carotid artery causing acute neck pain. A comprehensive history, high degree of suspicion, awareness of this entity and its transient nature among the medical practitioners are essential to make the diagnosis and alleviate patient's anxiety. Imaging plays a pivotal role in arriving at the diagnosis by excluding other sinister pathologies and also for follow-up after anti-inflammatory medications.

Abbreviations

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| TIPIC | Transient perivascular inflammation of carotid artery |
| MRI | Magnetic Resonance Imaging |
| USG | Ultrasonography |
| CT | Computed tomography |
| CCA | Common carotid artery |

Acknowledgements

Not applicable.

Author contributions

PG had diagnosed and followed up the case. PG and ID referred various journals and articles, studied extensively about the case, compiled data and prepared the manuscript. Both authors read and approved the final manuscript.

Funding

Not applicable.

Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Informed written consent was obtained from the patient and hospital for publication of the clinical details and radiology images.

Competing interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Received: 14 April 2023 Accepted: 18 September 2023

Published online: 25 September 2023

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