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Case Report
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TIPIC Syndrome – A Rare Cause of Neck Pain Case Report

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Abstract

Neck pain is a clinical complaint that encompasses a range of causes across different body systems. TIPIC (Transient Perivascular Inflammation of the Carotid artery) syndrome should be considered as a rare cause of neck pain. We present a normally fit and well 60-year-old man who presented with gradual onset unilateral neck pain which did not fit the presentation of any common illnesses. MRI was performed to assess the cause of neck pain, and TIPIC syndrome was subsequently diagnosed. TIPIC syndrome should be considered once other causes of neck pain are ruled out, to give clarification, reassurance and context to patients.

Keywords: TIPIC syndrome; Carotidynia; Transient perivascular inflammation of the carotid artery; Neck pain

Introduction

TIPIC syndrome is a relatively new diagnosis, which defines a group of patients with neck pain with some consistent features. It is useful to consider this as a diagnosis when other important causes are ruled out, to give both clinicians and patients understanding and reassurance. TIPIC syndrome or transient perivascular inflammation of the carotid artery syndrome was a term first adopted by Lecler et al. in 2017 [1]. This was a retrospective study based on 47 patients found to have similar clinical presentation and prognosis, all with the radiological finding of "thickened wall and infiltration of perivascular fat (or adventitia) involving the carotid bifurcation" Historically, TIPIC syndrome was known as Carotidynia, or Fay syndrome, first described by Fay in 1927 as "atypical radiating pain in the neck with tenderness over carotid bifurcation". This was dismissed as a symptom rather than a clinical entity until clarified in 2017 by Lecler et al. Based on this study, it is said that TIPIC syndrome composes of 2.8% of all acute neck pain [1].

Since the characterization study of TIPIC syndrome by Lecler in 2017, many case reports have been published. In 2022, Mecieli et al. published a further retrospective study based on 72 patients. The two retrospective cohorts both report median age of 48 at onset, with female to male ratio of around 1.5 to 1.

These studies collectively report symptoms of TIPIC syndrome which include acute neck tenderness, reported to be 92% unilateral, but in rare cases can be bilateral [2]. The pain may radiate to the face, ear, or jaw [3-5]. There may be pain on swallowing [6]. Fever may be present. Local swelling or lymph nodes may be present on examination. Although subsequent case studies have not reported neurological symptoms, Lecler reported 17% of patients in his study had neurological symptoms. These include extrinsic ipsilateral oculomotor cranial nerve palsy, contralateral transient motor deficiency, and peripheral facial palsy [1].

Here we present a patient who was seen in the ENT clinic for neck pain, who demonstrates the possible clinical presentation of TIPIC syndrome.

Case Presentation

A 60-year-old man who is normally fit and well, without long-term medical conditions was seen in the ENT clinic. He presented with 4 weeks history of gradual onset, deep left cheek pain. Gradually this cheek pain disappeared, and the pain moved to the left level 2 area on the neck, with pain radiating to the left post auricular area. The pain was intermittent, particularly noticeable on some neck movements, and sometimes on swallowing. The pain was not severe enough to warrant oral analgesia. The pain in the cheek was almost gone at this clinic review.

He described no other symptoms, including no sore throat, fever, dysphagia, voice changes, or shortness of breath. He had no otologic symptoms and no vertigo.

The patient had a normal examination of the oral cavity and oropharynx. A nasal endoscopy

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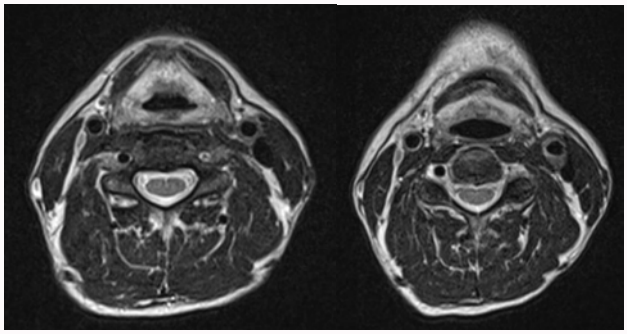

Figure 1: MRI showing thickening of the distal left common carotid artery and carotid bifurcation.

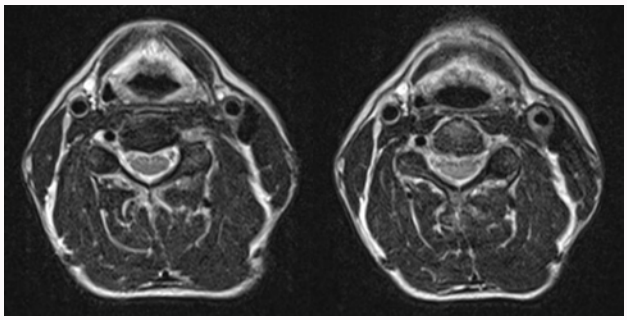

Figure 2: Three months follow up MRI showing similar appearances of distal left common carotid artery and carotid bifurcation.

was performed, and this was unremarkable. He had tenderness on palpation over the carotid pulse in the left level 2 area. No masses or lumps were palpable around this area or elsewhere in the neck. Ear examination was normal.

Due to the unusual unilateral symptom, an MRI of the neck was performed. MRI showed subtle thickening of the distal left common carotid artery and proximal external carotid artery, with T2 signal and enhancement following contrast. A small lymph node sits adjacent to it. Findings were suggestive of vasculitis, or TIPIC syndrome (transient perivascular inflammation of the carotid artery) (Figure 1).

Blood tests performed at about 1 week after symptom onset showed normal full blood count and C reactive protein.

The patient was followed up over the phone approximately 2 weeks after the initial consult (approximately 6 weeks after the onset of symptoms), and his symptoms are almost completely gone.

A follow-up MRI at 3 months showed stable similar appearances. The patient is now asymptomatic (Figure 2).

Discussion

Lecler et al. [7], initial study which defined TIPIC syndrome reported that 89% of the patient has normal inflammatory markers. Some case reports report elevated ESR and CRP [8,9]. A proportion of case report though appeared to be consistent with this reported figure, reporting patients with normal inflammatory markers [3,5,10-13]. It is difficult to know that out patient had elevated inflammatory markers initially, however inflammatory markers were normal at 1 week whilst he was still symptomatic with positive radiological findings, it is likely that his inflammatory markers have never been elevated.

Various imaging modalities were included in case reports so far: CT, MRI and ultrasound. The most affected area is the carotid bulb or bifurcation, distal common carotid artery, and proximal internal carotid artery. Most often they are affected in the posterior and lateral aspects of the vessels. Atherosclerotic plaques and mild stenosis were visualized in some patients, however, no patient in either larger studies or in any case report found plaques causing hemodynamically significant changes [2,7]. CT angiogram often demonstrates contrast enhancement around the carotid artery. On MRI, increased contrast enhancement is seen on T1 image of the carotid wall and a high signal on T2 images [12]. PET-CT has been employed in some case reports, some of which showed localized uptake around the carotid whilst other cases did not demonstrate uptake [8,11,13]. All features of TIPIC syndrome can be identified on ultrasound alone, which is cost-effective.

It is important to remember that TIPIC is a self-limiting diagnosis that can only be diagnosed when other differentials are excluded. Although ultrasound is cost-effective in diagnosing TIPIC syndrome, other imaging modalities such as CT and MRI are valuable in excluding other serious conditions which may require urgent treatment.

TIPIC syndrome is not a type vasculitis, as the inflammation mostly involves the adventitia. This leads to eccentric thickening of the vessels and hence does not cause hemodynamically significant obstruction of flow [14].

Although it is reported that TIPIC syndrome can cause neurological symptoms, it is unclear how this is caused, especially when there is no report of ultrasound demonstrating hemodynamically significant plaques in the carotid artery. In practice, differentials such as stroke or carotid artery dissection need to be excluded first, as they require imminent treatment. Other differentials include infection in the oropharynx, oral cavity, salivary glands, migraines, head and neck cancers, cervical spondylitis, TMJ dysfunction, carotid body tumor, aneurysm, occlusion, and vasculitis.

There are no apparent long-term sequelae of TIPIC syndrome. Symptoms are reported to last for 14 days on average [2]. Lecler et al. [1], followed up the study population for a mean duration of 3 months. All patients made full recovery. The treatments most used are non-steroidal anti-inflammatory and steroids. Other treatment options include calcium channel blockers, selective serotonin reuptake inhibitors, triptans, colchicine and benzodiazepines [1,13,14]. Maggialetti report their patient improved without treatment, like our patient presented here [5]. 19% of patients had a clinical relapse with the same symptoms and imaging findings between 1 to 6 months [1,2]. 53% of patients had FU imaging, and all had decreased or no perivascular inflammation [1]. Out of 15 patients included in the case reports read for the research of this report, 10 had reported on follow-up imaging. The timing ranges from 7 days to 6 months after the initial presentation, all showing improvement or normal scans [3-5,8-11,15-17]. It is peculiar why our patient had no change on MRI 3 months later, though this appears to produce no symptoms for him.

Conclusion

TIPIC Syndrome can be considered as a potential diagnosis when patients present with symptoms including pain at the carotid bifurcation which is acute onset, perivascular infiltration on imaging, exclusion of other diagnoses, and improvement of symptoms and imaging findings within 2 weeks, either spontaneously or with

NSAID use.

The mechanism of TIPIC syndrome is not understood. More studies are required to further understand the pathophysiology of TIPIC syndrome. Due to its apparent benign clinical course, the diagnosis of TIPIC syndrome should be made after the exclusion of other important condition which requires urgent treatment.

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