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Case Report

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A case of jugular paraganglioma presented as temporomandibular joint disorder

Abstract

Background: Jugular paraganlioma is a slow-growing tumor originating from the paraganglion cells. The diagnosis of a jugular paraganglioma may be delayed due to its atypical symptoms. We report a case of jugular paraganglioma presented as temporomandibular joint (TMJ) disorder.

Clinical presentation: 29-years old woman with preliminary diagnosis probable atypical face pain. She experienced her pain for the first time 2 months prior to reference. Pain intensity was seven to eight out of ten points on Visual Analogue Scale, simple and combined analgetics and NSAIDs were ineffective. Pain was dull, aching, localized in the right temporal area with irradiation to the ipsilateral maxillar and periauricular areas. Pain was more intensive in the nighttime. Temporomandibular joint movement (maximum unassisted and assisted opening, right to left movements) also increased pain. Significant dilatation of right foramen jugularis with its right side destruction was detected by computed tomography with contrast. The radiological findings were consistent with those of a jugular paraganglioma.

Conclusion: This case confirms that in refractory headache and TMJ pain other reasons should be considered with rigorous investigation including neuroimaging.

Keywords: jugular paraganlioma

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Abbreviations: CT, computed tomography; TMJ, temporomandibular joint

Introduction

Jugular paraganlioma is a slow-growing tumor originating from the paraganglion cells. Its typical manifestations involve hearing loss, pulsatile tinnitus and lower cranial nerve deficits due to a mass effect. The diagnosis of a jugular paraganglioma may be delayed due to its atypical symptoms. We report a case of jugular paraganglioma presented as temporomandibular joint (TMJ) disorder.

Case presentation

29-years old woman was referred to University Headache Clinic with preliminary diagnosis probable atypical face pain. She experienced her pain for the first time 2 months prior to reference. Pain intensity was seven to eight out of ten points on Visual Analogue Scale, simple and combined analgetics and NSAIDs were ineffective. Her pain was dull, aching, localized in the right temporal area with irradiation to the ipsilateral maxillar and periauricular areas. Pain was more intensive in the nighttime. Temporomandibular joint movement (maximum unassisted and assisted opening, right to left movements) also increased pain. Before reference to our clinic the patient was treated with pregabalin (300 mg per day) and amitriptyline (50 mg per day) without significant improvement. On physical examination blood pressure was 120/70 mmHg, heart rate was 75 beats/min. Right temporal, maxillar, periauricular areas were visually without abnormalities. Temporomandibular joint mobility was limited; palpation of tight temporal and masseter muscles, the right pole was painful. Clicking noise during opening and closing movements was identified. Cutaneus allodynia was revealed in periauricular, temporal maxillare areas. Cranial nerves were intact.

Computed tomography (CT) with contrast (Ultravist 370, 100 ml) was performed (Figure 1). Significant dilatation of right foramen jugularis with its right side destruction was detected: the left one size was 8x7 mm, the right one was 12x14.6 mm. In fossa jugularis we revealed a soft tissue mass with clear contours, homogeneous, destroying right wall of foramen jugularis. Enhanced-CT examination showed a significant enhancing mass. The radiological findings were consistent with those of a jugular paraganglioma.[1]

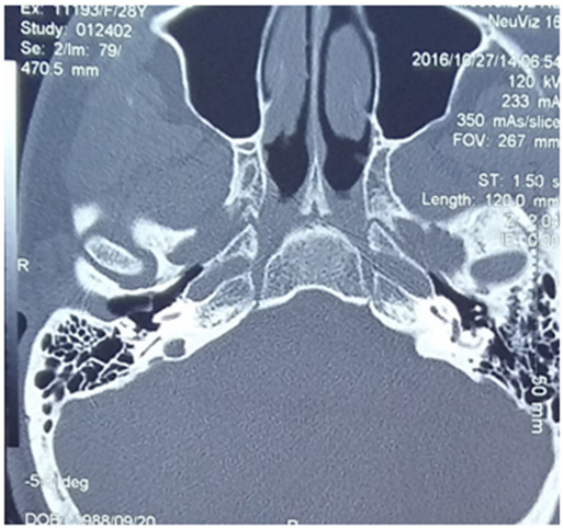

Figure 1 Results of computed tomography of the brain with contrast (Ultravist 370, 100 ml).

Discussion

Jugular paraganlioma is a rare benign tumor, which occurs in adults, typically between 40 and 60 years of age, more frequently among females. Typical symptoms are pulsatile tinnitus, hearing loss, hoarseness, dizziness. Headache occurs in 16% of the patients, otalgia - in 11%.[2] Jugular paraganglioma presented as temporomandibular

joint disorder is an extremely rare condition, we found only two similar cases in the literature.[3,4] The patient meets the criteria of temporomandibular arthralgia (International RDC/TMD Consortium Network and Orofacial Pain Special Interest Group)[5]: 1. Pain of joint origin that is affected by jaw movement, function, or parafunction and replication of this pain occurs with provocation testing of TMJ. 2. Pain in the jaw, temple, in the ear or in front of ear. 3. Pain is modified with jaw movements, function, or parafunction. 4. Conformation of pain location in the area of TMJ. 5. Report of familiar pain with palpation of the lateral pole and maximum unassisted and assisted opening, right to left movements.

We assume some possible reasons for atypical paraganglioma presentation in our patient. Mass effect on facial nerve can lead to subclinical facial asymmetry, laterality of proprioception in the orofacial muscles and TMJ dysfunction.[6] Paraganglioma rarely secrete various neuropeptide hormones, such as adrenocorticotropic hormone, and inflammatory cytokines, such as interleukin-6 (IL-6) inducing acute inflammatory reaction and pain.[7]

The patient was refractory to medication treatment: pain relievers, NSAIDs, pregabalin and amitriptyline were ineffective. This case confirms that in refractory headache and TMJ pain other reasons should be considered with rigorous investigation including neuroimaging.

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Conflicts of interest

None.

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