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Case Report

An unusual combined glomus vagale and jugular tumor: A case report

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ABSTRACT

Introduction: and importance: Head and neck paragangliomas are slowly growing benign tumors and they originate from specialized neural crest cells. We report an unusual combined glomus vagal and jugular tumor that was rarely described in the literature to the best of our knowledge.

Case presentation: A 51 years old female with no pathological history was presented to our ENT department with 6 months' history of a right latero cervical swelling gradually increasing in size associated with a swallowing difficulties and hoarseness. Preoperatively clinical examination had found vagal and hypoglossal nerve paralysis. Cervical CT scan and MRI had shown glomus jugular tumor. The patient underwent a surgical excision with severe swallowing difficulties and facial palsy in the immediate postoperative period with a mild recovery afterwards.

Clinical discussion: Paragangliomas of the mesotympanum and jugular foramen most commonly present as a vascular middle ear mass. The most common presenting symptom is pulsatile tinnitus occurring in 80% followed by hearing loss (60%). Dysfunction of cranial nerves traversing the jugular foramen may be commonly encountered with resultant abnormalities of speech, swallowing and airway function. Vagal paragangliomas are the least common of the three primary craniocervical paragangliomas. The most common presenting sign is the presence of a painless neck mass accompanied occasionally by dysphagia and hoarseness. The association of both glomus vagal and jugular tumor is rarely described in the literature to the best of our knowledge.

Conclusion: Head and neck paragangliomas are slowly growing benign tumors and they originate from specialized neural crest cells. Vagal paragangliomas are the least common of the three primary craniocervical paragangliomas. The association of both glomus vagal and jugular tumor is rarely described in the literature to the best of our knowledge. The choice of treatment depends on the location, size, and also biologic activity of the tumor as well as the physical condition of the patient.

1. Introduction

Head and neck paragangliomas are slowly growing benign tumors and they originate from specialized neural crest cells. Paragangliomas (PGLs) are non-epithelial neuroendocrine neoplasms (NENs) [1] that derive from Para ganglia, derived from neural ridges, which are seen in close association with components of the sympathetic and parasympathetic nervous systems. These tumors can be divided into two groups: those related to the sympathetic system, located mainly in the posterior mediastinum and retro peritoneum, the most typical of which is the pheochromocytoma, and those associated with the parasympathetic system, located in the head and neck as well as in the anterior mediastinum [2]. Paragangliomas are rare, hyper vascular neoplasms with an overall incidence of 1 in 30,000–100,000, head and neck paragangliomas arise, in order of decreasing frequency, from the carotid body, jugular bulb, vagal nerve, the tympanic branch of the glossopharyngeal or auricular branch of vagal nerve, and the cervical sympathetic chain [3]. The carotid paraganglioma is the most common head and neck paraganglioma, although the incidence is low [4]. The most common presenting sign for carotid paraganglioma is that of a painless cervical mass. The presence of a bruit over the mass is uncommonly noticed but, when present, suggests significant compression of the artery. Bulging of the oropharyngeal wall occurs in approximately 10% of tumors [5].

Paragangliomas of the mesotympanum (glomus tympanicum) and jugular foramen (glomus jugulare) most commonly present as a vascular

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middle ear mass. The most common presenting symptom is pulsatile tinnitus occurring in 80% followed by hearing loss (60%) [6]. Dysfunction of cranial nerves traversing the jugular foramen may be commonly encountered with resultant abnormalities of speech, swallowing and airway function.

Vagal paragangliomas are the least common of the three primary craniocervical paragangliomas. The most common presenting sign is the presence of a painless neck mass accompanied occasionally by dysphagia and hoarseness.Cranial nerve dysfunction occurs more frequently than with carotid paraganglioma but less than that of glomus jugular tumors.

We report an unusual combined glomus vagal and jugular tumor that was rarely described in the literature to the best of our knowledge.

2. Clinical presentation

This work has been reported in line with the SCARE 2020 criteria (7). researchregistry7207.

A 51 years old female with no pathological history (drug, genetic or psychosocial history), was presented to our ENT department with 6 months' history of a right latero cervical swelling gradually increasing in size associated with swallowing dysfunction and hoarseness. On the clinical examination, the mass was found to be painless, soft, pulsating and semifixed. During the otoscopic examination, no pulsatile erythematous lesion of vascular appearance was noted behind the eardrum. Oral examination had found a bulge of the right lateral pharyngeal wall pushing inside the tonsillar space. Furthermore, right cranial nerve paralysis was present. The vagal and hypoglossal nerve were paralyzed. Nasofibroscope had shown a mild paresis of the right vocal fold.

Cervical Ultrasound examination highlighted the presence of soft tissue mass, measuring 18.5 × 25 mm, hyper vascular, located in the right jugular vein. Cervical CT scan had found a strongly enhancing right parapharyngeal mass extending into jugular foramen without any bone destruction, measuring 35x28x51mm (Fig. 1). Cervical MRI had shown a mass of the right internal jugular vein in hyposignal T1, hypersignal T2 strongly enhancing with a salt and pepper appearance related to a glomus jugular tumor, with lateral sinus stasis. We performed a temporal bone CT scan to eliminate an associated glomus tympanicum tumor and to evaluate the skull base radiological anatomy. The glomus jugular tumor was classified stage C according to the Fish classification.

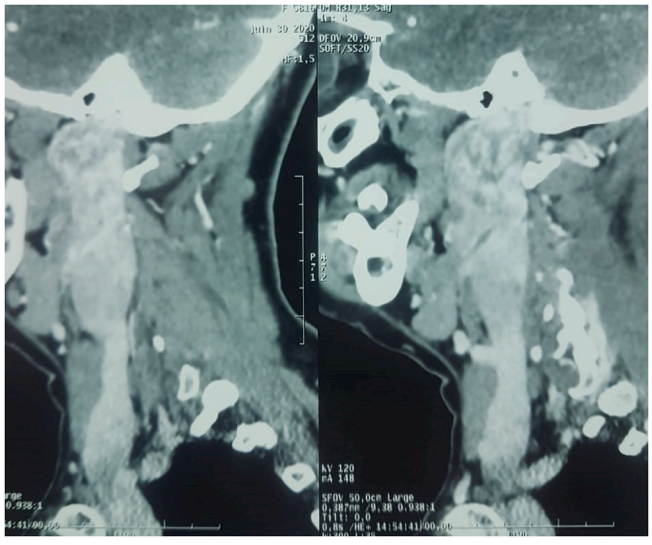

Fig. 1. Cervical CT scan showing a strongly enhancing mass of the right internal jugulare vein extending into jugular foramen without any bone destruction.

The methoxylated derivatives urinary catecholamines were in the normal range. Arteriography revealed a tumor blush in the right jugular vein reflecting the hyper vascular nature of the lesion which received its blood supply from the right ascending pharyngeal and sublingual arteries. We performed a cervical embolization, with ONYX 16, of the pharyngeal ascending artery under local anesthesia 24h before surgery. No complications occurred.

Two months after the first presentation in the consultation, the patient underwent a surgical excision by two professors, under general anesthesia through transmatoid + transcervical approach. During the operation, we had found 2 masses, a jugular one as well as a vagal one with external carotid artery invasion and right hypoglossal nerve lesion (Fig. 2).

The external carotid artery was ligated to allow easier control of hemorrhage. As a result, we did external carotid, vagale in addition to a hypoglossal nerve sacrifice. Facial nerve was isolated, skeletonized until the stylomastoid foramen. Next, it was transposed and completely mobilized from its fallopian canal, it was gently retracted and placed anteriorly along the parotid gland, which has been freed from its attachments to the sternocleidomastoid muscle. We performed a mastoidectomy to reach the sigmoid sinus, the infralabyrinthine bone overlying the jugular bulb and sigmoid sinus was removed, with the mastoid tip to allow adequate visualization of the jugular bulb. The sigmoid sinus was occluded. With this double approach, we did an entire excision of the tumor (Fig. 3).

Histopathological examination showed a vagal and internal jugular paraganglioma with well-defined nests of cuboidal cells separated by highly vascularized fibrous septa.

In the immediate postoperative period, the patient had developed severe swallowing difficulties, that required a nasogastric tube, with a complete right facial palsy. She stayed in hospital for two weeks until she had a mild recovery from her dysphasia. After 2 months of rehabilitation, she went to stage 3 of facial nerve paralysis according to House- Brackmann classification, without any swallowing abnormalities.

3. Discussion

Head and neck paragangliomas are slowly growing benign tumors and they originate from specialized neural crest cells. The association of both glomus vagal and jugular tumor is rarely described in the literature to the best of our knowledge. There was one case reported in the study of HUGH F. BILLER et al. conducted in 1989 gathering 18 patients [8].

Doppler ultrasound has limited function for glomus tumors and is only useful to assess and make differential diagnosis of carotid paraganglioma from lateral cervical masses [9,10]. The principle in glomus

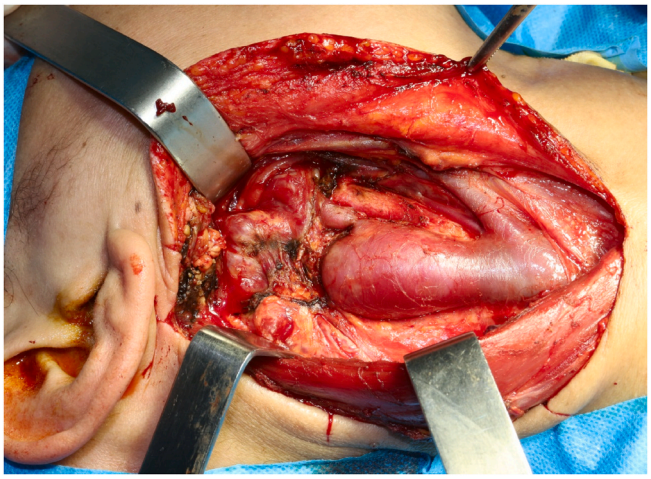

Fig. 2. Combined glomus vagal and jugular.

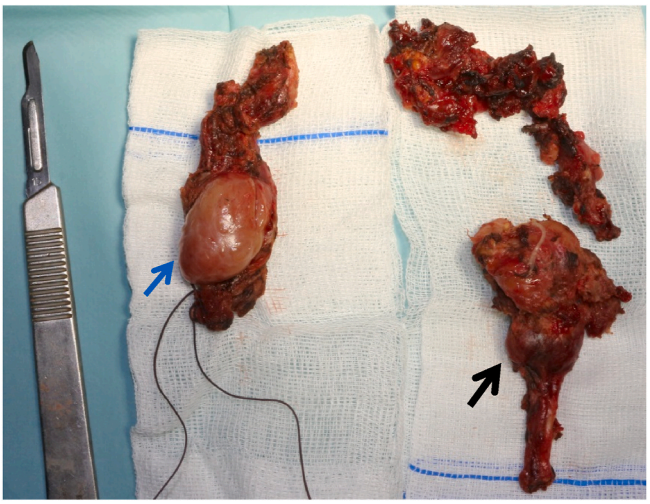

Fig. 3. Glomus jugular inside the internal jugular vein (black arrow) next to the glomus vagale (blue arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

tumor assessment is fine-slice computed tomography (CT) scanning [11]. CT scanning will show enhancing mass and bone windows can reveal skull base erosion along with intracranial extension. Magnetic resonance imaging (MRI) represents the essential imaging technique for evaluation and characterization of head and neck glomus tumor. In some of the T2-weighted scans, areas of bright signal intensity were present, giving a salt-and-pepper appearance to the lesion. MRI is more sensitive than CT scanning for tumors involving skull base and vascular structures [9]. MRI angiography is an imaging modality, which shows arterial and venous structures in a non-invasive fashion. Telischi et al. [12] considered octreotide scintigraphy a reliable test to detect both primary, and concurrent glomus tumors, and may be entirely helpful in the planning of surgery. However, the use of this imaging method in glomus tumors is limited. A minority of glomus tumors (1–3%) demonstrate clinical evidence of hormonal activity [13]. Urine vanillylmandelic acid levels were investigated in patients with signs of functioning tumor and a pathologically increased level was detected.

The extension of the tympanojugular paraganglioma can be assessed through Fisch classification. Class A is a glomus arising along the tympanic plexus on promontory. The class B invades the hypotympanum with intact cortical bone over the jugular bulb. The class C1 erodes the carotid foramen, the class C2 destroys the vertical carotid canal and the class C3 involves the horizontal carotid canal, while the class C4 invades the foramen lacerum and cavernous sinus. In class De, there is an intracranial but extradural extension ($De_1 < 2$ cm, $De_2 > 2$ cm, according to the displacement of the dura). In the class Di, there is an intracranial with an intradural extension ($Di_1 < 2$ cm, Di_2: between 2 and 4 cm, $Di_3 > 4$ cm, according to depth of invasion into the posterior cranial fossa) [14].

The main treatment options for paragangliomas include embolization, surgery, fractioned radiation (RT), radiosurgery, and a wait-and-scan strategy, to be used alone or in combination.

The first embolization of a glomus jugular tumor was described by Hekster and colleagues in 1973 [15]. This involved a transfemoral approach, selective catheterization of the arterial branches feeding the tumor. Since that time, there have been many reports on the benefits of preoperative embolization, with the resulting reduction of intra-operative bleeding, tumor size, as well as the operative time, in addition to facilitating the tumor resection. Indeed, the more invasive forms such as Fisch class C and D tumors, preoperative embolization allows for a more favorable surgical approach and increased probability of complete and uncomplicated tumor excision. [16] The recommended delay between embolization and surgery should be at least 2–3 days. This will give the edema time to subside but should not be prolonged beyond 2 weeks—otherwise, embolized feeding vessels may reopen, or new feeding arteries may be recruited [17,18].

The subject of pre-operative embolization in managing vagal paraganglioma represents an issue of controversy. Urquhart et al. [19], have pointed out the possibility of incurring neurologic complications as a consequence of embolization and have concluded that there appears to be no surgical advantage to performing this procedure. In the Jefferson series, cited by Miller et al. [20], no neurologic complications were encountered with these embolized patients. The investigators felt strongly that absence of bleeding, as a result of embolization, contributed to a safer dissection around neighboring cranial nerves, thus limiting the potential for morbidity associated with nerve injury.

Fractioned radiation (RT) and stereotactic radiosurgery (SRS) produce long-term, durable HNPGL control, providing excellent tumor control over 2–119 months of follow-up and causing no treatment complications. These therapies are clearly highly-efficacious, safe alternatives to surgery and are best suited for patients with large carotid paraganglioma, jugular paraganglioma, vagal paraganglioma, or multifocal HNPGL who have a high likelihood of operative morbidity to lower cranial nerves or contraindications to surgery (eg, medical comorbidities) [21].

Class C and D on Fisch classification of temporal bone jugular glomus usually require an infratemporal approach. If intradural extension is present, Fisch $Di_{1/2}$, the tumor should be resected in a two-stage, team-approach procedure involving neurosurgeons. This therapeutic option is not available for Fisch Di_3, where palliative radiotherapy is proposed [14].

According to the study of Joshua D. Smith et al. conducted from 2000 to 2017 gathering 194 patients with head and neck paragangliomas, they have established an algorithm of management of these tumors. For old asymptomatic patients, with co morbidities, small tumor and SDHx mutation (low malignancy risk), watchful waiting protocol was employed with biannual CT/MRI. In the opposite case or If there is a tumor growth, with new symptoms or cranial nerve palsies, then treatment is indicated (surgery, RT, stereotactic radiosurgery (SRS)). For glomus vagale and jugular tumor associated with pain, compression and cranial nerve palsies, surgery was indicated with pre-op tumor embolization. In case of subtotal resection, RT and SRS are recommended as adjuvant treatment. For painless glomus vagale and jugular tumor with no cranial nerve paralysis, RT and SRS are suggested without surgical approach [3].

Even if surgical resection is the recommended treatment for paraganglioma, with the main goal of excising the neoplasm and preventing local advancement, for patients with bilateral mass, surgery may not be performed all at once to avoid serious complications including cranial nerve dysfunction: approximately 10% of the cases had nerve injury of the hypoglossal, glossopharyngeal, recurrent-laryngeal, or spinal accessory nerves, or involvement of the sympathetic chain [22].

Facial nerve palsies and swallowing difficulty have been reported in the postoperative period [23,24]. Dysphagia remains a major contributor to post-operative morbidity in this type of surgery. It was the same case in our patient. In the study of J.E. Fenton et al. [25] in the management of dysphagia in jugular foramen surgery gathering 61 patients, twenty-two developed a permanent cranial nerve palsy after surgery including 14 vagal deficits and eight cases of hypoglossal injury. All patients had moderate to severe dysphagia post-operatively but only two required permanent enteral feeding (via a gastrostomy).Refinements in the management of the facial nerve during the infratemporal fossa approach have allowed improved facial nerve function in the immediate post-operative period [26]. Facial nerve preservation has been further enhanced through the use of intra operative facial nerve monitoring [27]. Exclusion of patients with prior radiotherapy further enhances preservation of facial function during mobilization. The poorer results in patients with pre-operative irradiation is likely secondary to fibrotic changes within the neural microcirculation making mobilization less

tolerable. In our case report, our patient hadn't undergone irradiation prior to the surgery however, no facial nerve monitoring was used (the nerve monitoring system was not functional).

To overcome the surgical morbidity associated with vagal deficits multiple attempts of preserving vagal anatomy at surgery or reconstruction of cut nerve were reported in the literature without constant favorable results. If hoarseness is the only bothersome symptom, medialization thyroplasty is the proposed form of rehabilitation. Primary medialization thyroplasty by silastic implant has also been tried for acute glottis incompetence to avoid need for tracheostomy and its associated morbidity.

Acute onset dysphagia, in selected patients, can be managed with conservative management till opposite vagal nerve compensates [28]. Hypoglossal nerve injury results in paralysis of the ipsilateral tongue and hemiatrophy of the tongue within a few months. Swallowing therapy will be then, required to prevent aspiration and is based on educating the patient to direct the bolus to the functioning side. More significant, persistent swallowing and aspiration issues may require feeding via tracheostomy and gastrostomy tubes [29].

In histopathological examination, these tumors are usually composed of solid nests known as "zellballen" of round to oval or elongated cells with abundant granular amphophilic or basophilic cytoplasm, within a vascular stroma, but they may also have acidophilic cytoplasm. There may be nuclear atypia but usually mitoses are scarce, and there is no necrosis. Immunohistochemistry plays a key role in confirming the diagnosis. However, many pathologists believe that the only stains required are chromogranin, synaptophysin and S100 [30].

4. Conclusion

Head and neck paragangliomas are slowly growing benign tumors and they originate from specialized neural crest cells. Vagal paragangliomas are the least common of the three primary craniocervical paragangliomas. The association of both glomus vagal and jugular tumor is rarely described in the literature to the best of our knowledge. The choice of treatment depends on the location, size, and also biologic activity of the tumor as well as the physical condition of the patient.

Patient perspective

The patient should share her perspective on the treatment she had received maybe one month after surgery and rehabilitation.

Sources of funding

None.

Ethical approval

No needed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution statement

Hammouda yassir, bushra abdulhakeem, khadija elbouhmadi: study concept data collection, writing the paper, and making the revision of the paper following the reviewers instruction.

Youssef oukessou, regragui meryem, sami rouadi, reda abada, mohamed roubal, mohamed mahtar: reviewing and validating the manuscript credibility.

Research registration

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Provenance and peer review

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Declaration of competing interest

None.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102918.

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