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Case Report Open Access

Preoperative Onyx Embolization of Glomus Jugulare Tumor Complicated by Surgical Displacement of Embolic Material: Case Report

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Abstract

Onyx Liquid Embolic System (LES) embolization of highly vascular lesions is becoming more frequently used as an adjuvant therapy prior to surgical resection. The authors present a case of a successful pre-operative embolization of a glomus jugulare tumor using Onyx LES. The patient returned for scheduled operative resection one week later. Immediately following the surgery, the patient experienced new onset left hemiplegia. Full radiological workup revealed a fragment of Onyx LES occluding the middle cerebral artery (MCA). All indications are that the fragment became dislodged during the operation. To our knowledge, there have been no other reported cases where complications of tumor embolization with Onyx LES occurred during surgical resection.

Keywords: Onyx; Glomus jugular; Pre-operative embolization; Stroke

Introduction

Onyx LES was FDA approved in 2005 for the embolization of arteriovenous malformations (AVM) prior to surgical resection. It has been used more recently as adjuvant therapy prior to definitive surgical resection in the embolization of paragangliomas [1], meningiomas [2], vertebral hemangiomas [3], juvenile nasal angiofibromas [4], among other tumors. Onyx LES is becoming more frequently used because it has been reported to offer particular advantages including more extensive tumor devascularization, the use of fewer arterial catheterizations, and greater safety during catheter withdrawal [4]. In this paper, we describe a rare complication of surgical displacement of Onyx LES embolic material from a pre-operatively embolized glomus jugulare tumor. A fragment of embolic material was surgically displaced from an extracranial position during operative resection of a glomus tumor into the intracranial circulation resulting in an ischemic stroke and hemiplegia.

Case Report

History and examination

This 65-year old woman presented after 3 years of progressive vertigo, nausea, and pulsatile tinnitus in the right ear. Over the 7 months previous to presentation she had lost much of the hearing in her right ear. On examination she had a subtle right sided facial droop and complained of peri-oral numbness along the right side of her face. There was mild weakness of the right trapezius. Her hearing was markedly diminished on the right side and there was a red bulging mass visible behind the right tympanic membrane.

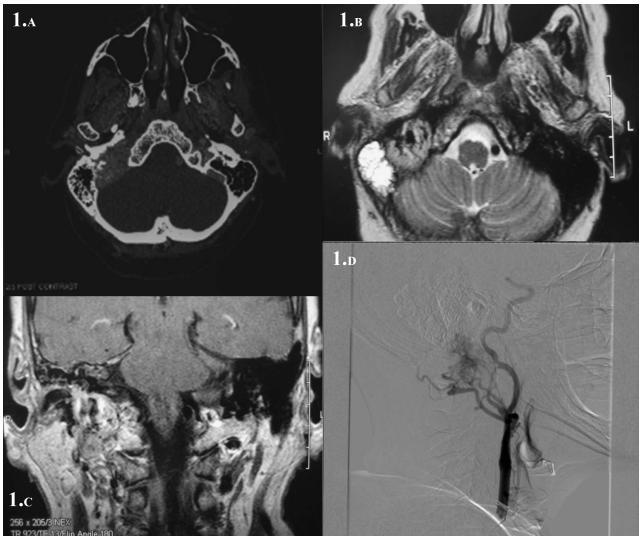

Figure 1: CT, MR, digital subtraction angiography obtained at first presentation and as part of pre-operative evaluation. A. Axial CT image showing moth-eaten appearance erosion of bone around the jugular foramen and intensely enhancing mass. B. Axial T2-weighted MR image demonstrating T2 hyperintense lesion with scattered hypointense foci giving a "salt and pepper" appearance. C. Coronal T1-weighted with contrast MR image showing enhancing mass extending from the jugular foramen to the level of the C3 ring. D. Lateral view of common carotid artery injection demonstrating enlarged occipital and ascending pharyngeal branches of the external carotid artery feeding the tumor.

CT imaging of her head showed a moth eaten pattern of erosion expanding the jugular foramen with an intensely enhancing homogenous mass lesion (Figure 1A). Magnetic resonance (MR) imaging showed a mass with low signal intensity on T1 that enhanced intensely following intravenous contrast administration (Figure 1B). The mass extended inferiorly to the level of C3. The tumor had high signal intensity on T2 weighted images with the classic "salt and pepper appearance" of multiple areas of high and low intensity signals representing fast and slow vascular flows. Digital subtraction angiography was performed to define the vascular anatomy feeding the tumor (Figure 1C and 1D). The tumor was fed primarily from the ascending pharyngeal artery and the occipital artery branches of the external carotid artery. There was also contribution from enlarged muscular branches of the vertebral artery on the right side.

Embolization

To facilitate operative resection, the decision was made to embolize the mass prior to surgery (Figure 2). Distal occipital and ascending pharyngeal branches of the external carotid artery presented challenges for subselective catheterization even after multiple microwire/catheter combinations were attempted. A decision was made to place coils in the occipital artery for distal protection. Subsequently, using Onyx LES, the glomus tumor was embolized through the microcatheter positioned within the occipital artery. Another microcatheter was then positioned within the external carotid artery in the region of the ascending pharyngeal artery and additional tumor embolization was performed. The Onyx embolic material was allowed to reflux to within a few centimeters of the common carotid bifurcation. The embolization continued until there was no evidence of persistent angiographic filling. The contributing branches from the vertebral artery were considered minor and were left untreated. The patient was discharged to home on post-embolization day 2 without any new neurologic deficits.

She returned 5 days later for her scheduled surgical resection. The operation was performed by a multidisciplinary team of neuro-otologists and neurosurgeons. An infratemporal fossa transmastoid approach combined with posterior fossa craniectomy along with cervical dissection was performed for optimal exposure and resection of the tumor. The sigmoid sinus was ligated at the jugular bulb during the procedure. Blood loss was well controlled throughout the procedure which lasted about 12 hours. Intra-operative neuromonitoring was not used during the case.

On awakening from anesthesia, the patient was discovered to have a new left sided hemiplegia. She was sent for immediate CT imaging of her head where a hyperdense fragment was identified in the right sylvian fissure (Figure 3). CT angiography confirmed that the embolic material was within the right middle cerebral artery at the branch point of the bifurcation. The duration of time the MCA had been occluded was unknown, but was likely extended given the length of her surgery. There was early evidence of hypodensity on the CT scan suggesting a prolonged period of ischemia. No endovascular intervention was attempted and she was started on aspirin and Plavix for anti-platelet therapy.

Subsequent MR imaging was obtained demonstrating a new cerebral infarction in the region of the right lentiform nucleus and posterior internal capsule (Figure 3D). Her physical exam remained compromised with a dense left hemiplegia and the patient was discharged to acute care rehabilitation.

Figure 2: Digital subtraction angiography obtained during pre-operative embolization procedure. A. Lateral view showing Onyx embolic material within the occipital artery. Coils were placed prior to onyx embolization to provide distal protection. B. AP view showing embolized ascending pharyngeal artery with back fill into the external carotid artery. C. AP view of common carotid injection showing minimal residual tumor filling from anterior circulation and onyx embolic material occluding the external carotid just distal to the superior thyroid artery branch point.

Discussion

There have been significant advancements in the last thirty years with regard to the surgical management of glomus jugulare tumors (paragangliomas). Improvements in care during the pre-operative period have helped reduce the risks associated with surgical resection. The highly vascular nature of the tumor can make hemostasis challenging when operative resection is indicated. Limiting intraoperative blood loss allows for a more complete resection of the tumor and helps minimize the risk of injury to surrounding cranial nerves [5-7]. Several techniques have been established for devascularizing glomus jugulare tumors by embolization and occlusion of tumor feeding vessels. Embolization facilitates surgical resection by reducing the size of the tumor, thereby relieving some of the mechanical burden of the resection, as well as by reducing the intraoperative blood loss [5,8].

The methods for preoperative embolization of glomus tumors have evolved over the last several decades. In 1980, Schick, et al. reported

successful embolization of a paraganglioma with coils and other particulate agents via transarterial catheterization [9]. This approach persisted for several years and proved to be efficacious for significantly decreasing blood loss during resection [5,10] by occlusion of tumor feeding vessels.

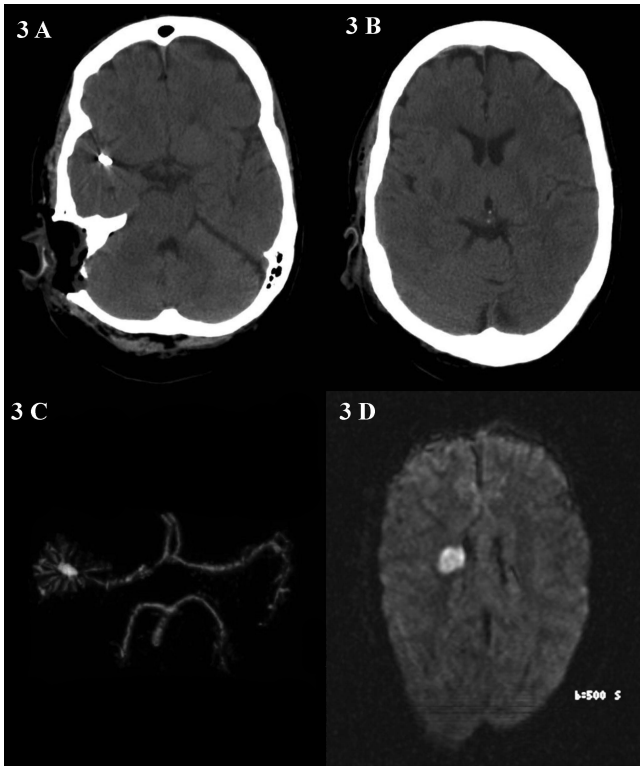

Figure 3: Imaging obtained following surgical resection of the glomus jugulare showing occlusion of the right middle cerebral artery by a displaced fragment of onyx material. A. Axial CT image obtained immediately following resection of tumor showing hyperdense metallic fragment lodged within the right sylvian fissure. B. Higher cut of axial CT showing early hypodense lesion in MCA distribution. C. 3-dimensional reconstruction of CT angiogram with shine artifact of onyx fragment occluding the MCA at the M2 bifurcation point. D. Diffusion weighted MR image obtained on the third post-operative day confirming new cerebral infarct in region of right lentiform nucleus and posterior internal capsule.

There were possible complications observed from transarterial catheterization which included stroke caused by migration of embolic material into the intracranial circulation during the embolization procedure [7]. This led to the development of direct percutaneous embolization where the embolic material is injected percutaneously into the neoplastic tissue rather than by way of the arterial supply. Studies describing percutaneous embolization, beginning with Casasco in 1994, showed it to be a more reliable method for tumor devascularization when compared to the transarterial technique [11-14]. Nevertheless, the possibility of embolic stroke still persisted with tumor embolization by the percutaneous approach as well [12].

In addition to the developments of embolization technique for delivering occluding agents into tumor vasculature, continued research has focused on the types of materials used to devascularize these paragangliomas. Common embolizing agents include cyanoacrylate-type glues, such as n-butyl cyanoacrylate (NBCA). A new substance introduced in the past several years which is gaining more widespread use is Onyx Liquid Embolic System (LES), an ethylene vinyl copolymer in suspension with tantalum powder in dimethyl-sulfoxide (DMSO), which is delivered by microcatheter. Onyx LES has gained popularity because of its reported advantages in achieving more extensive tumor devascularization, using fewer arterial catheterizations, and doing so with a greater safety profile during catheter withdrawal [4].

Regardless of the technique of delivery or the occluding agent used, it has been traditionally understood that the risks associated with preoperative tumor devascularization were directly attributable to the embolization procedure itself. As noted, the most dangerous of these risks is embolic stroke from dislodged or improperly placed embolic material that is swept into the cerebral vasculature and occludes a downstream artery during the embolization procedure. In contrast, the case outlined here involved the successful embolization of a glomus jugulare tumor using transarterial microcatheterization with Onyx LES in the preoperative period. The patient was discharged home post-embolization day 2 with no existing cardiac or neurological complications and returned 5 days later in the same condition. Immediately following the surgery, the patient experienced new onset left hemiplegia. Full radiological workup revealed a fragment of Onyx LES occluding the middle cerebral artery (MCA). All indications are that the fragment became dislodged during the operation.

We reason that manipulation of the tumor during surgical resection likely caused a fragment of onyx to become dislodged from the proximal portion of the external carotid artery close to its junction with the common carotid artery where it was subsequently swept downstream into the internal carotid artery until becoming wedged within the ipsilateral MCA thus causing an ischemic infarct. Other potential routes through external carotid arterial collaterals to the internal carotid circulation or through an arteriovenous shunt are unlikely because the volume of the onyx cast seems larger than the requisite vasculature for such routes [15]. The intact neurologic status of the patient post-embolization makes it unlikely that the embolus had occurred during the embolization procedure. Furthermore, there were no episodes of hemodynamic instability, hypotension, or hemoconcentration during the operation that might have caused ischemia in an already compromised vascular distribution were the embolus to have been present prior to surgical resection.

Embolization of hypervascular head and neck tumors with onyx is largely considered to be a safe procedure. Rangel-Castilla et al. reported the successful onyx embolization of 100 consecutive head, neck, and spinal tumors including meningiomas, paragangliogliomas, juvenile nasal angiofibromas, hemangiomas and various metastatic lesions [16]. Seventy-nine of the patients underwent post-embolization surgical resections. Complications occurred in only 10% of patients and were generally minor including cranial nerve palsies (n=2), groin hematomas (n=3), extravasation of onyx around the tumor (n=3), and myocardial infarction in the peri-embolization period (n=2). There were no incidents of death or stroke.

There are several important lessons to be gleaned from this case. The first is that Onyx-18 (6% ethylene vinyl alcohol (EVOH) copolymer) and Onyx-34 (8% EVOH copolymer) are soft and pliable, possibly predisposing them to displacement. Secondly, the surgeon needs to be aware of the possibility for displacement of the embolic

material during surgery. This extends to adapting the surgical technique in such areas as tying-off vessels and the relative gentleness of dissection techniques. Additionally, interventionalists should be careful to avoid placement of Onyx LES near critical vessel branches (e.g. ICA) where displacement of embolic material may occur. Lastly, when Onyx LES is involved, interventionalists should consider use of additional embolizing agents (e.g. coils) to prevent displacement. Communication between the surgeon and endovascular interventionalist is of utmost importance in order to define therapeutic plans including which vascular pedicles to target, associated risks of embolization, and the risks of operative approaches to resection.

This case study presents a new and unique challenge in the surgical management of difficult and highly vascularized tumors. To our knowledge, there have been no other reported cases where complications of tumor embolization with Onyx LES occurred during surgical resection.

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