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Case Report

Preoperative Endovascular Embolization of Orbital Solitary Fibrous Tumor With 500-700 Micron Tris-Acryl Gelatin Microspheres

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Abstract

The reported experience with preoperative embolization of solid orbital tumors is scarce. Herein, we present a case of a large and hypervascular orbital solitary fibrous tumor (SFT) in which 500-700 µm tris-acryl gelatin microspheres (TAGM) were used for preoperative embolization. A 41-year-old man presented with severe proptosis, palpable mass, restrictive myopathy, exposure keratopathy, and compressive optic neuropathy in the right orbit. Magnetic resonance imaging showed a 65x35x35 mm, diffusely contrast-enhanced tumor in the superior orbit, extending to the apex, and multiple intratumoral vascular flow voids. A diagnosis of SFT was made by incisional biopsy. Endovascular tumor embolization was performed with 500-700 µm TAGM. Two days later, the tumor was entirely removed with minimal bleeding. No embolization- or surgery-related complications and tumor recurrence or metastasis developed during the 42-month postoperative follow-up.

Keywords: Orbit, solitary fibrous tumor, surgical treatment, tumor embolization, tris-acryl gelatin microsphere

Introduction

Solitary fibrous tumors (SFTs), relatively rare orbital lesions, are often hypervascular.[1] Large, hypervascular SFTs extending to the apex of the orbit constitute a serious surgical challenge. Due to limited access to the apical-posterior part of the tumor, it can be difficult to control intraoperative bleeding and the tumor may be only partially removed, leading to serious postoperative morbidities. Preoperative endovascular tumor embolization is a rarely used method for solid orbital tumors. Herein, we present the removal of a large orbital SFT after embolizing with 500-700 µm tris-acryl gelatin microspheres (TAGM). This report adheres to the tenets of the Declaration of Helsinki, and written informed consent was obtained for publication of patient information and images.

Case Report

A 41-year-old man presented with increasing proptosis for over 1 year, pain, and blurred vision in his right eye (Figure 1A).

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An immobile, hard mass was palpated in the superolateral orbit, pushing the globe inferomedially. There were large subcutaneous vessels in the upper eyelid. External ocular motility was restricted in all directions, and an afferent pupillary defect was present. Visual acuity was counting fingers at 2 meters in the right eye and 20/20 in the left eye. There was exposure keratopathy, diffuse optic disc swelling, and choroidal folds in the right eye.

Magnetic resonance imaging showed a well-defined, large (65x35x35 mm) soft tissue mass with lobulated contours in the right orbit (both extra- and intraconal), filling the posterior orbit. The lesion was isointense to gray matter on T1- and T2-weighted images and avidly enhanced on T1-weighted postcontrast imaging (Figure 2A-C). There were multiple vascular signal voids in and adjacent to the lesion (Figure 2B). Expansile remodeling of the adjacent orbital bony walls was noted on computed tomography images (Figure 2D). An incisional biopsy revealed an extrapleural SFT that demonstrated strong immunoreactivity for CD34 and STAT6. A light microscope showed uniform oval to spindle cells organized into whorls. There was a typical patternless pattern, without significant mitotic activity, necrosis, or cellular atypia (Figure 3).

With the patient under intravenous sedation, tumor embolization was performed using 500-700 µm TAGM (Embosphere, Merit Medical Systems Inc., Utah, USA). Carotid angiography via the femoral artery showed a hypervascular mass fed by the internal maxillary and middle meningeal branches of the right external carotid artery (Figure 2E, F). Both feeding arteries were catheterized with a microguidewire (Hybrid, Balt, Montmorency, France) and Rebar microcatheter (Medtronic, Irvine USA). Under fluoroscopic control, a mixture of contrast media and microspherical particles was slowly injected until the passage of the contrast agent through the feeding arteries ceased (Figure 2G). There were no complications after the procedure, except for orbital pain responsive to intravenous paracetamol.

Two days after the embolization, the tumor was entirely removed by a combined superior and lateral orbitotomy through an extended upper eyelid crease incision (Figure 1B). The lesion had caused significant fat atrophy in the upper orbit and was tightly adhered to the surrounding tissues despite having a pseudocapsule. Bleeding from the tumor could be easily controlled during surgery. After tumor removal, redundant upper and lower eyelid tissues were trimmed, and the lateral canthus was reconstructed (Figure 1C). The tumor did not recur during 42 months of follow-up. At the last examination, superior sulcus depression, ptosis, and exotropia were noted (Figure 1D). Extraocular motility was normal in all directions, except a mild restriction in upgaze. Visual acuity was 20/100 in the right eye.

Discussion

SFTs constitute up to 1-3% of all orbital masses, and the main treatment is surgical resection.[2] If not completely removed, SFTs may recur and exhibit malignant transformation. These complications are reported to occur in up to 37% and 12% of orbital cases, respectively, and can manifest after many years.[3,4] Tumor diameter (>3 cm) may be a significant risk factor for local recurrence and distant metastasis.[5] Large SFTs extending

Figure 1. Preoperative (A), intraoperative (B and C), and 28-month postoperative (D) photographs of the patient with orbital solitary fibrous tumor

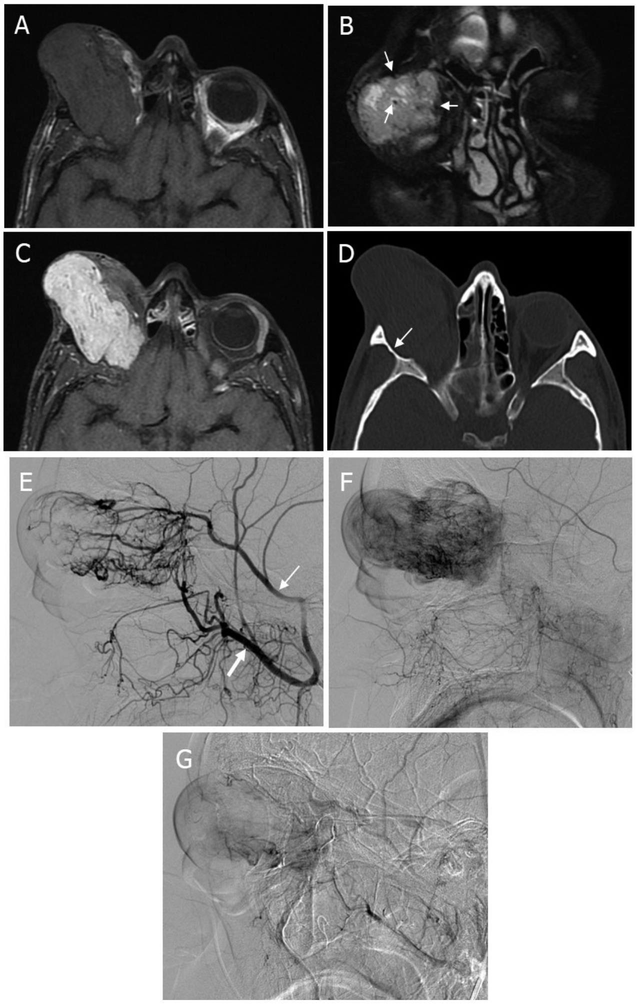

Figure 2. Magnetic resonance imaging (A-C) shows a large soft tissue mass with a well-defined, lobulated contour. A) The lesion was isointense to gray matter on axial T1-weighted images. B) Coronal fat-suppressed T2-weighted image shows multiple intratumoral vascular flow voids (arrows). C) On axial contrast-enhanced fat-suppressed T1-weighted image, the lesion enhanced strongly. D) Axial computed tomography image demonstrates remodeling of the lateral orbital wall due to mass effect (arrow). E) Microcatheterization angiography shows a large and highly hypervascular mass fed through the internal maxillary (thick arrow) and middle meningeal (thin arrow) arteries, F) a strong tumor blush in the late arterial phase, and G) a significant decrease in the tumor blush after embolization with microspherical particles

to the apex of the orbit may be complicated by bleeding and incomplete removal due to the difficulty in accessing the tumor pedicle. Preoperative embolization can reduce tumor size, surgical bleeding, and operative time. Liquid, particles, or coils can be used for embolization. Each material has physical properties that make it advantageous for specific indications.[6]

Liquid embolizers are typically used in aneurysms, arteriovenous malformations, and arteriovenous fistulas, while coils are preferred for occlusion of large vessels.[6,7] Particulate embolizers (polyvinyl alcohol or TAGM) are most often used for tumors. TAGM are approved specifically for uterine and prostate tumors. These particles can be injected with microcatheters smaller than their maximum diameter due to their flexible, compressible, and non-aggregate structure. They do not cause clogging and sticking in or around the catheter orifice. Although other embolizing agents cause vascular occlusion where they exit the catheter, microspherical particles can penetrate deeper. Because of these physical properties, TAGM may be preferred over polyvinyl alcohol.[6] TAGM adhere to the vascular endothelium and can remain for 4 weeks without causing severe inflammation and degeneration.[8] However, due to possible revascularization, surgical procedures should be performed within a few days after embolization. The embolizing microspheres may cause an allergic reaction because they contain porcine gelatin.

Preoperative embolization is a rarely utilized technique for solid tumors in the orbit. A recent review found that only 3 of the 275 patients with SFT in the literature were managed with preoperative embolization.[5] In these cases, liquid agents such as n-butyl-2-cyanoacrylate glue and ethylene vinyl alcohol copolymer (Onyx®) were used for embolization.[9,10,11] The mechanical coil was used in another case in the Japanese literature.[12] Recently, two cases of SFT in which Onyx was used by intraoperative intratumoral injection or via preoperative transophthalmic arterial route were reported.[13,14] The tumor was removed after piecemeal resection in the first case and after enucleation in the other.[13,14] Endovascular tumor embolization with particulate material has been previously reported in only one case of SFT.[15] In this case, an extraconal SFT in the superoanterior-mid orbit was embolized with 150-300 µm TAGM injected through the distal ophthalmic artery.

In a large and hypervascular tumor in the posterior orbit, ideally all major feeding vessels should be occluded for effective

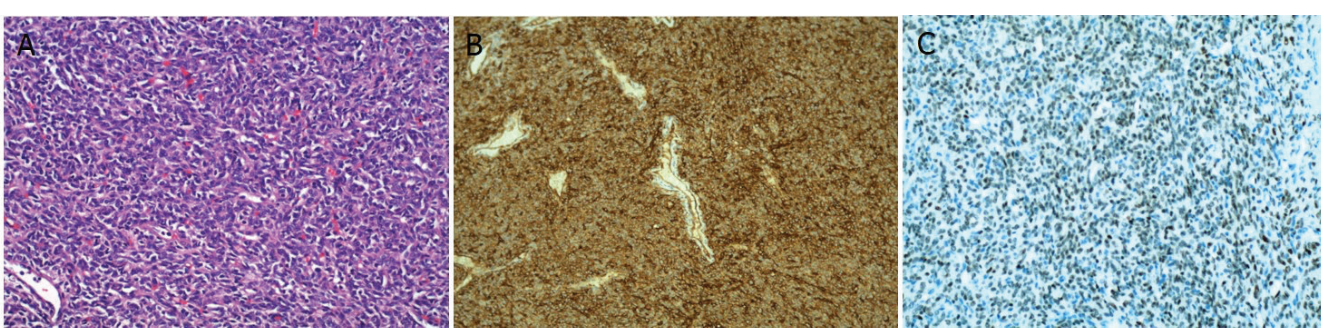

Figure 3. A) Histologically, the tumor is composed of uniform oval to spindle cells. The tumor cells exhibited diffuse CD34 positivity (B) and nuclear STAT6 expression (C)

devascularization due to the dense collateral anastomoses within the tumor. However, the embolization procedure can have serious complications. In a recent case, embolization of the ophthalmic artery with Onyx resulted in sudden, extreme orbital pain, severe conjunctival congestion, and oculocardiac bradycardia requiring immediate treatment, as well as permanent vision loss.[14] Intercarotid anastomoses, common arteries that supply both the tumor and normal structures, the caliber of feeding vessels, and potential reflux of embolic agent into the parent vessel are all factors that need to be considered before a decision to embolize.[7,16]

TAGM are available in six different size ranges (40-120 µm to 900-1200 µm). Particle sizing is critical for efficacy and complications.[8] While smaller particles are able to penetrate more distally into tumor capillary beds, they can also enter the extratumoral circulation through collateral anastomoses.[14] Small particles (>300 µm) may cause more frequent complications in intracranial meningiomas.[17] In the present case, a large orbital SFT was safely embolized with 500-700 µm TAGM and could be completely surgically excised. This material may be suitable for endovascular embolization in orbital solid tumors.

Ethics

Informed Consent: It was obtained.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practice: B.Y., B.H., Z.Y., U.Y., Concept: B.Y., B.H., Design: B.Y., B.H., Data Collection or Processing: B.Y., B.H., Ö.E.M., Z.Y., U.Y., Analysis or Interpretation: B.Y., B.H., Ö.E.M., Z.Y., U.Y., Literature Search: B.Y., Ö.E.M., Writing: B.Y., Ö.E.M., Z.Y.

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