Case Report

Concomitant Orbital Cavernous Hemangioma and Solitary Fibrous Tumor

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Abstract

Cavernous hemangiomas are the most frequent tumors in the orbital region. Solitary fibrous tumors are uncommon neoplasms found in different locations, including the orbit. We present a 50 year old man with coincidence of unilateral orbital cavernous hemangioma and solitary fibrous tumor. Computed tomography was performed which showed two intraconal solid masses in the right orbit positioned relatively near each other antro-postriorly. Tumors were excised using lateral orbitotomy method and sent for histopathologic examination. Our case shows the coincidence of a solitary fibrous tumor adjacent to cavernous hemangioma so this combination is possible and should be considered.

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Introduction

Solitary fibrous tumors (SFTs) have a mesenchymal origin [1] and are relatively rare neoplasms found in visceral tissue particularly in pleura [2- 4], but rarely develop in other locations including peritoneum, mediastinum, liver, pericardium, retroperitoneum and other anatomical structures [5, 6]. Very rarely, SFTs develop in the eye orbit mostly originating in the fourth decade of life [1]. These tumors are extremely rare among children, but they have been reported in some cases in the age range of 5 months to 16 years [5, 7].

The clinical observations of patients with SFTs consist of swelling, asymmetry, slowly progressive proptosis over an average of two years, loss of eyeball mobility and patients might complain from pain, diplopia and visual loss as well [5, 8]. Diagnosis of SFTs is made by immunochemical analysis, therefore positive CD 34 is the main clinical test to validate the primary observations [1]. Although the surgical excision of the whole tumor is considered as a gold standard, it can be difficult due to friability and high vascularity [9]. In addition using radiotherapy can be helpful in reducing the tumor size before and after the surgery [9].

Le et al. reviewed the key features of orbital SFT (OSFT) in 84 involved cases; the incidence of OSFT according to sex was estimated at 54 % and 45.7 % among males and females, respectively. Although the painless proptosis is considered as the most prevalent clinical presentation among these patients, ptosis might be rarely induced (3.5 %) [10]. The most common location for the tumor is the supero-medial orbital wall [10]. It has been reported that 74 % of OSFTs are indolent and are not aggressive [10].

We present a 50 year old man with coincidence of unilateral orbital cavernous hemangioma and solitary fibrous tumor.

Case Report

A 50-years-old Iraqi man presented with priorbital edema and proptosis and gradually increasing mild pain in the right eye (since many years ago). Uncorrected visual acuity was 20/200 in the right eye and 20/20 in the left eye, but corrected visual acuity with cyclorefraction (+2.50 - 0.50 × 45°) was 20/25 in the right eye.

On external examination the right eye had 4 mm proptosis, but ocular motility was normal and no afferent pupillary defect was detected. Slit lamp examination was normal in both eyes, the right eye's intraocular pressure was 17 mmHg and the left eye's intraocular pressure was 13 mmHg. Fundoscopic examination was normal in both eyes and optic disc appearance was pink with sharp margins.

Computed tomography (CT) was performed. On CT two axial and intraconal separate solid masses in the right orbit positioned relatively near each other antro-postriorly were observed.

The lateral orbitotomy method was planned in order to obtain the best access to remove the tumor and the excised tumor was sent for the histopathologic examination. The lateral orbital wall was dissected from the bone with blunt periosteal elevation. The lateral periorbita was dissected posteriorly to the posterior one fourth of the orbit, exposing the lateral orbital wall. After excising the anterolateral orbital wall, removal of the posterior wall was completed. A periorbital incision was made, avoiding the lateral rectus muscle. Handheld retraction was used to retract the globe anteriorly and to retract the orbital soft tissues. When the tumor was identified, it was dissected using blunt techniques with cotton-tipped applicators and microdissection instruments. A cryoprobe with special elongated orbital tip was placed on

the lesion to provide traction. The cryoprobe was sequentially replaced as the tumor was rolled out. The process continued until the tumor was freed up entirely and could be removed. After achieving complete hemostasis, closure was performed.

Macroscopic examination of the histopathologic observations revealed an ovoid grayish soft tissue (2 × 1.5 × 1cm) with hemorrhagic background accompanied by two irregular grayish-brown soft tissues (1.5 × 1.5 cm).

In addition, microscopic evaluation of histopathologic observations showed a benign neoplastic lesion characterized by patternless architecture of spindle cells, branching hemangiopericytoma like vessels, and keloidal type collagen deposition (Figure 1).

with cavernous hemangioma accompanied with solitary fibrous tumor.

Discussion

Cavernous hemangiomas are the most frequently found primary tumors in the orbital region. They normally appear in adults [11, 12]. Solitary fibrous tumors are uncommon neoplasms found in visceral tissue particularly in pleura [2-4]. These tumors are also rarely found in the orbit [2-4]. Although orbital cavernous hemangiomas are typically unilateral, some reports have reported bilateral cases [13-15]. Ali et al., [1] presented a 22 year old man with proptotic eyeball and downward displacement due to OSFT which caused ocular movement restriction. Tumor was excit-

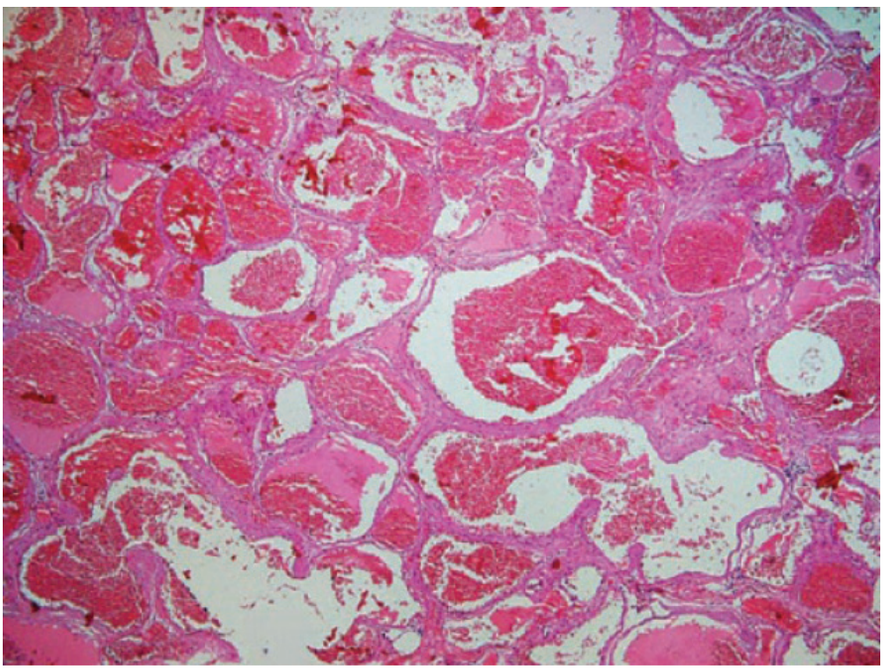

Figure 1: Evaluation of histopathologic observations showed a benignneoplastic lesion characterized by patternless architecture of spindlecells, branching hemangiopericytoma like vessels, and keloidal type collagen deposition.

Also a benign neoplastic lesion composed of spindle cells having elongated nuclei with focal mild nuclear atypia was observed. On immunohistochemistrical study spindle cells showed strongly positive CD 34 and CD 99 (Figure 2). Therefore the diagnosis was compatible

ed completely along with the overlying capsule. There was no evidence of tumor recurrence after 6 months of follow up examinations. Le et al. have reported four patients with unilateral OSFTs inducing painless proptosis, facial deformity, globe displacement and also diplopia [10]. The tumor

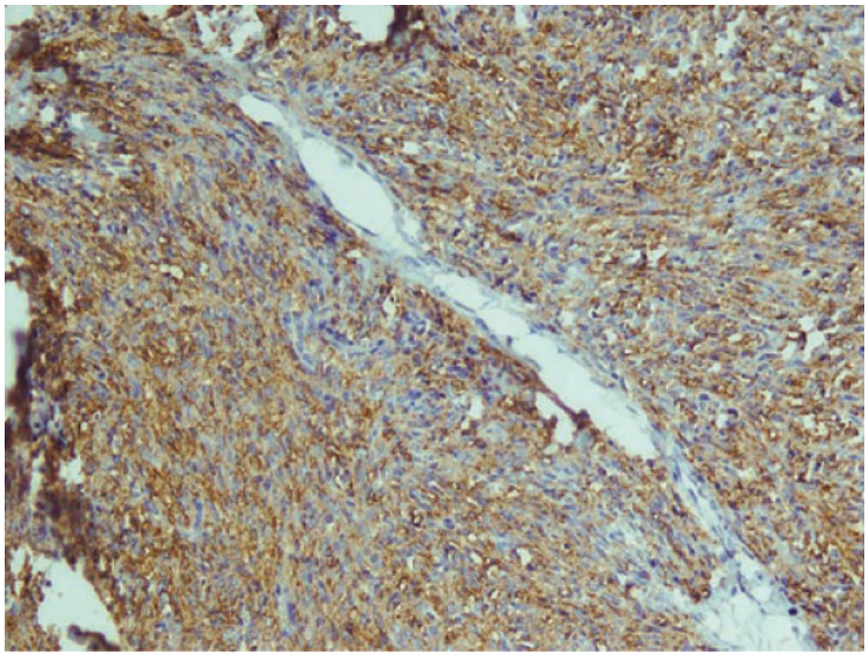

Figure 2: In immunohistochimestrical study spindle cells show strongly positive CD 34 and CD 99.

was removed completely with medial and lateral orbitotomy [10]. The pathological diagnosis of a solitary fibrous tumor, in comparison to other mesenchymal tumors such as hemangiopricytoma, can be difficult [16]. Bcl-2 and CD 99 immunohistochemistriy were used to distinguish a solitary fibrous tumor from a hemangiopriocytoma because a solitary fibrous tumor is positive for Bcl-2 and CD 99 but these markers are negative for hemangiopricytoma [16]. To our knowledge only one case of splenic hemangiopricytoma adjacent to cavernous hemangioma of colon have been reported in the English literature [17], but no case of solitary fibrous tumor and cavernous hemangioma has been previously reported.

Conclusion

Our case shows the coincidence of a solitary fibrous tumor adjacent to cavernous hemangioma so this combination is possible and should be considered.

References

1. Ali MJ, Honavar SG, Naik MN, Vemuganti GK. Orbital solitary fibrous tumor: A clinicopathologic correlation and review of literature. Oman J Ophthalmol. 2011;4(3):147-9.

2. Furusato E, Valenzuela IA, Fanburg-Smith JC, Auerbach A, Furusato B, Cameron JD, et al. Orbital solitary fibrous tumor: encompassing terminology for hemangiopericytoma, giant cell angiofibroma, and fibrous histiocytoma of the orbit: reappraisal of 41 cases. Hum Pathol. 2011;42(1):120-8.

3. Ing EB, Kennerdell JS, Olson PR, Ogino S, Rothfus WE. Solitary fibrous tumor of the orbit. Ophthal Plast Reconstr Surg. 1998;14(1):57-61.

4. Cerdá-Nicolás M, Löpez-Gines C, Gil-Benso R, Benito R, Pellin A, Ruiz-Saurí A, et al. Solitary fibrous tumor of the orbit: morphological, cytogenetic and molecular features. Neuropathology. 2006;26(6):557-63.

5. Meyer TN, Matos BH, Oliveira LR, Mendonça AT. Report of a case of solitary fibrous tumour of the orbit. Oral Maxillofac Surg. 2013;17(3):225-7.

6. Harmouch A, Chefchaouni MC, Maher M, Sefiani S. Solitary fibrous tumor of the orbit: report of two cases and review of literature. J Fr Ophtalmol. 2011;34(2):133-7. (Article in French)

7. Ribeiro SF, Chahud F, Cruz AA. Orbital hemangiopericytoma/solitary fibrous tumor in childhood. Ophthal Plast Reconstr Surg. 2012;28(3):e58-60.

8. Farah-Klibi F, Ferchichi L, Zaïri I, Rammeh S, Adouani A, Jilani SB, et al. Lipomatous hemangiopericytoma (adipocytic variant of solitary fibrous tumor) of the orbit. A case report with review of the literature. Pathologica. 2006;98(6):645-8.

9. Shinder R, Jackson TL, Araujo D, Prieto VG, Guadagnolo BA, Esmaeli B. Preoperative radiation therapy in the management of recurrent orbital hemangiopericytoma. Ophthal Plast Reconstr Surg. 2011;27(5):e126-8.

10. Le CP, Jones S, Valenzuela AA. Orbital solitary fibrous tumor: a case series with review of the literature. Orbit. 2014;33(2):145-51.

11. Herter T, Bennefeld H, Brandt M. Orbital cavernous hemangiomas. Neurosurg Rev. 1988;11(2):143-7.

12. Anand R, Deria K, Sharma P, Narula M, Garg R. Extraconal cavernous hemangioma of orbit. Indian J Radiol. 2008;1(8):310-12.

13. Aletaha M, Erfanian-Salim R, Bagheri A, Salour H, Abrishami M. Bilateral orbital cavernous hemangiomas J Ophthalmic Vis Res. 2010;5(1):65-7.

14. Fries PD, Char DH. Bilateral orbital cavernous haemangiomas. Br J Ophthalmol. 1988;72(11):871-3.

15. Sullivan TJ, Aylward GW, Wright JE, Moseley IF, Garner A. Bilateral multiple cavernous haemangiomas of the orbit. Br J Ophthalmol. 1992;76(10):627-9.

16. Kitahata Y, Yokoyama S, Takifuji K, Hotta T, Matsuda K, Tominaga T, et al. Hemangiopericytoma in the sacrococcygeal space: a Case Report. J Med Case Reports. 2010;14;4:8.

17. Yilmazlar T, Kirdak T, Yerci O, Adim SB, Kanat O, Manavoglu O. Splenic hemangiopericytoma and serosal cavernous hemangiomatosis of the adjacent colon. World J Gastroenterol. 2005;11(26):4111-3.

Footnotes and Financial Disclosures

Conflict of Interest:

The authors declare no conflict of interest with the subject matter of the present manuscript.