Case Report

Subconjunctival Myolipoma Confirmed with Immunohistochemical Analysis: A Case Report

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Abstract

Purpose: To report the clinicopathological features of a rare case of subconjunctival myolipoma and its treatment results.

Case Report: A 17-year-old female patient referred to our center with a white-pink mass in her left upper bulbar conjunctiva. The lesion extended to the forniceal conjunctiva. The patient had otherwise normal complete ocular examinations and underwent complete surgical excision of the mass due to cosmetic concerns. The tumor was examined with light–microscopy, following hematoxylin and eosin (H&E), Masson-trichrome, and immunohistochemical (IHC) staining.

A definite diagnosis of subconjunctival myolipoma was acheived following the pathological assessment. Six months postoperatively, no tumor recurrence was noted, and ocular examinations were within normal limits.

Conclusion: To date, only a few cases of orbital myolipoma have been reported in the literature. To the best of our knowledge, the current manuscript is the first report of subconjunctival myolipoma in the English literature. A straightforward diagnosis is possible through pathological assessment of the excised tumor and surgical treatment shows good results.

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Introduction

Myolipoma of the soft tissue, which was first described in 1991 is commonly reported in the abdominal cavity and retroperitoneum[1, 2] and is defined as a benign tumor consisting of mature adipose cells mixed with differentiated smooth muscle cells [3]. This is an exceedingly rare tumor with less than 50 reported cases in the literature and female preponderance [4]. In 1998, the first case of orbital myolipoma was reported in a 67-year-old female as left lower medial eyelid involvement and in 2003 the second case was reported, who was a 41-year-old Japanese female with right extraconal mass. In both patients, the diagnosis of myolipoma was confirmed using histological examination, immunohistochemical (IHC) staining, and transmission electron microscopy assessment [5, 6].

To the best of our knowledge, the present manuscript presents the first rep[ort of subconjunctival myolipoma in English literature.

Case report

A 17-year-old immunocompetent female presented with a mass of white-pink color in the left upper bulbar conjunctiva, extended and limited to the forniceal conjunctiva (Figures 1A and 1B). The patient had noted this mass six months prior to the visit, which was asymptomatic, and due to cosmetic concerns, had decided to excise the lesion. This subconjunctival mass was not mobile and was covered with intact white-pink color conjunctiva with no associated features including hair follicle, tenderness or sentinel vessels.

Patient's corrected distance visual acuity (CDVA) using Snellen visual acuity chart was 20/20 in both eyes and refraction was within normal limits. Intraocular pressure was 15 mmHg in both eyes and extraocular movements were also normal. The posterior and anterior segment examinations were unremarkable. A systematic review of the patient's history and physical examination was normal.

Following a written informed consent from the patient's father, surgical excision was performed using non-touch technique with 2 mm uninvolved margin under topical anesthesia with 0.05 % tetracaine (Anestocaine, Sina Darou, Tehran, Iran) and 2.5 % phenylephrine (Sina Darou, Tehran, Iran). The lesion was localized and totally excised then stored in a sealed container consisting of 10 % formalin and sent for routine pathology examination. Postoperatively eye drop of 0.5 % chloramphenicol (Sina-Darou,

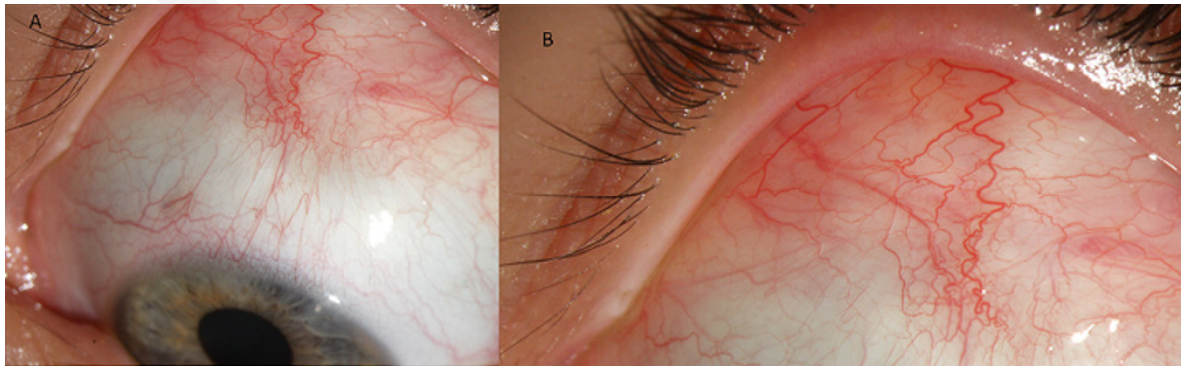

Figure 1: Slit photographs showing the clinical appearance of the white-pink color mass in the left upper bulbar conjunctiva (A), extended toward the forniceal conjunctiva (B)

Tehran, Iran) every six hours and 0.1 % fluorometholone (Fluocort 0.1 %, Sina-Darou, Tehran, Iran) every eight hours was prescribed for one month for her left eye.

The macroscopic examination revealed two soft fragments of creamy-gray tissue, covered by mucosa measuring 8 mm × 7 mm in surface and 4 mm in thickness. The specimen was wrapped in a filter paper and totally submitted for histopathological processing.

The paraffin sections were stained using hematoxylin and eosin (H&E) staining, and following the inspection of the pathological features of a tumor, it was decided to use further stains, including Masson-trichrome and immunohistochemical (IHC) staining. We used the following antibodies for IHC staining: HMB45 (Biogenex, San Ramon, CA; Ready-to-Use Antibody), actin (Biogenex, San Ramon, CA; Ready-to-Use Antibody), desmin (Biogenex, San Ramon, CA; Ready-to-Use Antibody), estrogen receptor (ER) (Biogenex, San Ramon, CA; Ready-to-Use Antibody), S100 protein (Biocare Medical, Walnut Creek, CA; Ready-to-Use Antibody) as well as smooth muscle actin (SMA) (Biocare Medical, Walnut Creek, CA; Ready-to-Use Antibody).

Light-microscopic examination of the H&E stained tissue showed mature adipose cells with no lipoblasts or floret-like giant cells, intermingled with spindle-shaped cells revealing a sieve-like pattern covered with conjunctival epithelium, which was not associated with atypical features, including apoptotic bodies, mitotic figures or necrosis (Figures 2A, 2B and 2C). Also, Masson-trichrome staining revealed muscle fibers in red color and collagen fibers in blue color (Figure 2D). The IHC staining revealed a positive reaction as brown staining with actin (Figure 3A), desmin (Figure 3B), and SMA (Figure 3) in muscle fibers and S100 protein

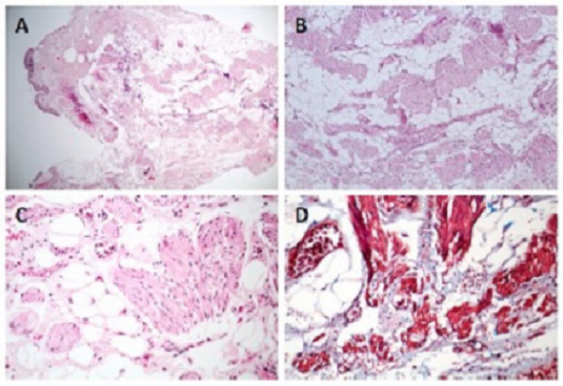

Figure 2: Subconjunctival myolipoma H&E staining: (A), (B) and (C) sections show subepithelial mature adipose tissue mixed with differentiated muscle fibers with band oval nuclei (H&E; original magnification ×40, ×100 and ×400, in A, B and C sections respectively). Masson-trichrome staining demonstrates muscle fibers in red color and collagen fibers in blue color (Masson-trichrome; original magnification ×400) (D)

(Figure 3) in mature adipocytes, yet the tumor was immunonegative for HMB45 (Figure 3) or ER (Figure 3). Considering the above-mentioned pathological findings, the definite pathological diagnosis of subconjunctival myolipoma was reached.

The examination of the patient, six months after the operation showed no signs of mass recurrence.

Discussion

Smooth muscle actin (SMA) expression by myofibroblasts is used for identification of these cells [7-9] and the presence of desmin has been observed in most smooth muscle cells and striated muscle cells [10]. In the current case, the smooth muscle component of the tumor was immunopositive for actin, desmin, and SMA, which was compatible with its muscle

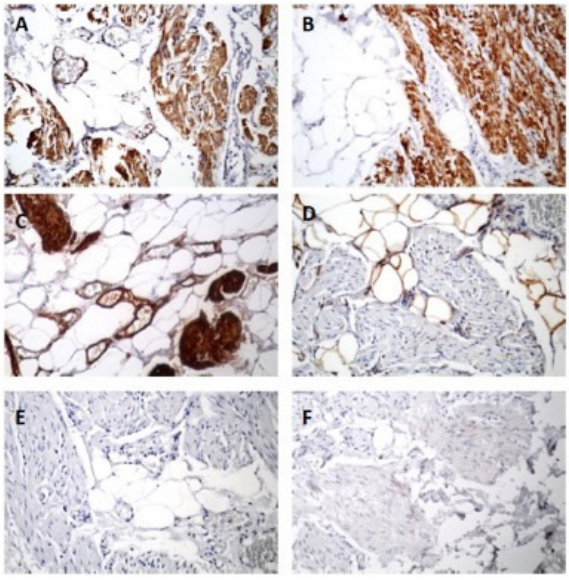

Figure 3: Subconjunctival myolipoma IHC staining: positive reaction as brown staining with actin (IHC; original magnification ×100) (A), desmin in muscle fibers (IHC; original magnification ×100) (B), SMA (IHC; original magnification × 400) (C), and brown staining with S100 in mature adipocytes (IHC; original magnification ×400) (D). Negative reaction is seen in staining with HMB45 (IHC; original magnification ×100) (E) and ER (IHC; original magnification ×100) (F)

element. This immunoreactivity was similar to Fukushima's myolipoma case series, who observed desmin immunoreactivity in 100 % and SMA immunoreactivity in 95 % of their cases [4]. Also, immunoreactivity for SMA and desmin in the first [5] and for SMA in the second [6] reported case of the orbital myolipoma was similar to our findings. Furthermore, studies have shown that mature lipocytes are immunoreactive for S 100 protein[11 12], which was observed in the lipid component of the tumor in the present case and confirmed the second component of the subconjunctival myolipoma.

Myoplipoma is an extremely rare tumor, and its presentation in ocular structures was first described in 1998 by Sharara et al.,[5] in a 67-year-old female with left lower lid mass, whose diagnosis was confirmed using H&E, Masson trichrome, IHC staining, and conventional transmission electron microscopic assessment. A strong positive reaction was noted for desmin and SMA in smooth muscle fibers and for Masson trichrome in non-striated intra-cytoplasmic filaments, which are comparable with findings of the present case report [5]. We did not perform a conventional transmission electron microscopy assessment due to the inaccessibility of the equipment in our center.

The second case of orbital myolipoma, which was presented as exophthalmos of the right eye, in a 41-year-old Japanese female, was described by Nagayama et al., [6] in 2003. This orbital myolipoma was located in the extraconal space of the right orbit, measuring 30 millimeters × 20 millimeters, as revealed in axial Tl-weighted magnetic resonance imaging. The excised tumor was examined pathologically using H&E, Masson trichrome, and IHC staining, together with conventional transmission electron microscopy assessment [6]. In IHC staining, a strong positive reaction was noted for SMA, yet none for Melan A, HMB45, and estrogen receptors [6]. The findings of the present investigation were in agreement with their case report. Despite a positive reaction with estrogen receptor, which was reported in 13 out of 15 myolipoma cases of the non-ocular site by Fukushima et al. , [4] we found no positive reaction with the estrogen receptor our case similar to the patient reported by Nagayama et al., [6] . This difference could be attributed to the difference in the location of the tumor in the orbital structures.

Similar to our case the majority of reported

myolipoma cases in the literature are females. In the case-series published by Fukushima et al.,[4] 94 % of subjects were female while both cases of orbital myolipoma previously reported were females [5 6]. However, a clear rationalization for this gender preponderance is yet to be determined.

Conclusion

To date, only a few cases of orbital myolipoma have been reported in the literature. To the best of our knowledge, the current manuscript is the first report of subconjunctival myolipoma in the English literature. A straightforward diagnosis is possible through pathological assessment of the excised tumor and surgical treatment shows good results.

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Footnotes and Financial Disclosures

Conflicts of Interest:

The authors have no conflict of interest with the subject matter of the present study.