

Conjunctival Metastasis from Leiomyosarcoma of Anterior Chest Wall

Sonal P Yadav^{1*}, Swapnil Patil¹, Anirudha Puntambekar², Rahul Deshpande¹

¹Department of Ophthalmic Plastic Surgery, Orbit and Ocular Oncology, PBMA's H V Desai Eye Hospital, Pune, Maharashtra, India

²Department of Histopathology, Ruby Hall Clinic, Pune, Maharashtra, India

Abstract

Leiomyosarcoma is the most common soft tissue sarcoma and is described as a malignant tumor of mesenchymal cells. It is rarely found as primary, secondary, or metastatic in origin in the eye and adnexa. To the best of our knowledge, a metastatic leiomyosarcoma involving the conjunctiva has not been reported in the literature. We report a case of a 35-years-old gentleman, with a prior history of leiomyosarcoma involving the anterior chest wall with pulmonary metastasis presenting to us with a rapidly increasing conjunctival mass involving his left eye. After a detailed clinical evaluation, the lesion was surgically excised. We discuss the clinical features and histopathology characteristics supported by immunohistochemistry analysis of this rare case of conjunctival metastasis from leiomyosarcoma with a review of metastatic tumors of the conjunctiva.

***Correspondence to:** Sonal P Yadav, Department of Ophthalmic Plastic Surgery, Orbit and Ocular Oncology, PBMA's H V Desai Eye Hospital, Pune, Maharashtra, India, E-mail: sonal.yadav@hvdeh.org

Citation: Yadav SP, Patil S, Puntambekar A, et al. (2020) Conjunctival Metastasis from Leiomyosarcoma of Anterior Chest Wall. *J Clin Oncol Ther*, Volume 2:2. 112. DOI: <https://doi.org/10.47275/2690-5663-112>.

Received: December 12, 2020; **Accepted:** December 22, 2020; **Published:** December 29, 2020

Introduction

Metastatic tumor to conjunctiva is a rare entity and it may often reflect poor prognosis due to an advanced state of the primary malignancy. They constitute <1% of all conjunctival tumors [1]. Leiomyosarcoma is a malignant tumor of mesenchymal cells with smooth muscle differentiation. Primary leiomyosarcoma originating from the eye and adnexa has been reported from conjunctiva, uvea, and orbit [2-4]. Metastatic leiomyosarcoma is extremely rare and has been described in orbit, eyelid, and choroid in literature [5-8]. To the best of our knowledge, leiomyosarcoma metastatic to conjunctiva has not been reported previously. We describe a case of a 35-years-old gentleman who presented to us with a rapidly growing nodule on his bulbar conjunctiva with a history of biopsy-proven left-sided anterior chest wall leiomyosarcoma with a review of metastatic tumors of the conjunctiva. The patient's consent was obtained for use of his clinical photographs for academic and research purposes. The case report adheres to the ethical principles outlined in the Declaration of Helsinki as amended in 2013.

Case Report

A 35-years-old gentleman presented to us with a rapidly increasing single reddish nodule on the nasal bulbar conjunctiva of the left eye noticed for 2 weeks. It measured 11x10x6mm, showed superficial congestion with intense intrinsic vascularity and episcleral fixity (Figure 1).

His best-corrected visual acuity was 20/20, both eyes with



Figure 1: External photograph of left eye showing elevated and vascular solitary conjunctival nodule on the nasal bulbar conjunctiva.

an unremarkable posterior segment in the left eye. The right eye examination was within normal limits. There was no palpable lymphadenopathy on the either side. On review of prior records, he was a diagnosed (biopsy-proven) case of leiomyosarcoma of the anterior chest wall with metastasis to lungs 8 years back and was treated with surgery, radiation, and adjuvant chemotherapy elsewhere. He was in remission as per the treating oncosurgeon until his current presentation. His routine blood investigations along with HIV serology was found to be within normal limits.

On detailed evaluation, the suspicion of metastatic nature of the solitary conjunctival nodule was considered. The nature of the ailment, pros, and cons of surgical treatment was discussed with the patient. He underwent wide excision of the conjunctival nodule with double freeze-thaw cryotherapy to the margins followed by fibrin glue-assisted surface reconstruction with amniotic membrane. Intra-operatively, the



nodule was found to be adherent to the underlying sclera. A controlled lamellar sclerectomy was performed to excise maximum tumor residue. On histopathology evaluation (H & E), the tumor was composed of spindle and pleomorphic cells with markedly atypical nuclei and brisk mitotic activity. (Figure 2A and 2B) Immunohistochemical (IHC) analysis revealed that the tumor cells were strongly positive diffusely for smooth muscle actin (SMA) (Figure 2C) and focally positive for Desmin (Figure 2D). Cytokeratin was absent. The features were consistent with metastatic leiomyosarcoma involving the conjunctiva.

The wound healing was satisfactory at the time of 6 weeks review with minimal residual scarring. (Figure 3A and B) As the base of the excised lesion was involved by the tumor, he was advised to undergo episcleral plaque brachytherapy to the tumor base along with systemic metastatic evaluation (Whole body PET CT scan) to look for progression at the primary site and/or additional distant metastasis. However, the patient has been non-compliant for further investigations and management. On the last contact, 9 months post-surgery, he is alive and stable.

Discussion

Metastatic tumors to conjunctiva present as rapidly growing, fleshy, pink, vascularised lesion often non-adherent to the underlying sclera [9]. The clinical presentation of the current case was as per the previous description except we encountered metastatic conjunctival nodule adherent to and involving underlying sclera. In absence of a prior documented history of anterior chest wall leiomyosarcoma,

the clinical differential diagnosis of the solitary conjunctival nodule in a young male would have included nodular episcleritis or nodular scleritis, foreign body granuloma, lymphoproliferative disease, or occasionally a mucoepidermoid variant of squamous cell carcinoma, myxoid, myogenic or histiocytic tumor [10]. The previous history and availability of histopathology diagnosis of primary malignancy along with immunohistochemistry aided histopathology evaluation of the present lesion helped to secure a rare diagnosis of metastatic conjunctival tumor from a leiomyosarcoma. On an extensive literature search on the PubMed database, we could not find a prior published case of metastatic conjunctival tumor from leiomyosarcoma making this the first reported case.

Metastasis to conjunctiva is notably rare. On PubMed search with keywords “conjunctiva/val” and “metastasis” in the English language, we found only 3 large series and few individual case reports describing metastasis to conjunctiva [1,9,11-33]. (Summarised in Table 1). The most common primary malignancy site was breast 14 cases followed by cutaneous melanoma 13 cases and lung 9 cases. Other reported primary malignancy sites include the larynx, gastric, renal, visceral Kaposi, parotid gland, utero-cervix, and vagina. Conjunctival metastasis was found on bulbar conjunctiva in the majority of reported cases followed by palpebral, tarsal, and forniceal sites. In most of the reported cases, the conjunctival metastasis was accompanied by rest of ocular structure involvement and/or wide-spread distant organ metastasis. Excision for conjunctival site coupled with definitive or palliative systemic

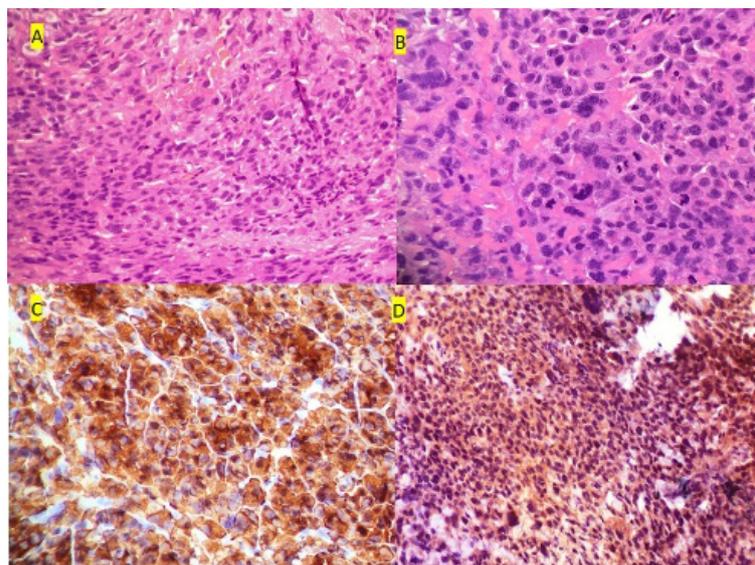


Figure 2: Histopathology shows tumor composed of spindle and pleomorphic cells with markedly atypical nuclei and brisk mitotic activity [Figure2A (H &E 200) and Figure2B (H & E 400)]; Immunohistochemistry (IHC) shows tumor cells were strongly positive diffusely for smooth muscle actin (SMA) (Figure2C) and focally positive for Desmin (Figure2D).



Figure 3: Slit lamp photograph of left eye showing the pre-operative (Figure 3A) appearance and post-operative appearance at 6 weeks of follow up (Figure 3B).



chemotherapy and/or radiotherapy was employed for management [1,9,11-33] (Table 1).

Leiomyosarcoma is the most common soft tissue sarcoma. It can arise from multiple sites including soft tissue (48%), skin (14%), gastrointestinal tract (7%), uterus (7%), and retroperitoneum (7%) [34]. Primary leiomyosarcoma of the chest wall is rare and reportedly accounts for approximately 1-4% of primary soft tissue sarcomas of the chest wall [35]. Distant metastasis from chest wall leiomyosarcoma can occur in the liver or lung via a hematogenous route [36]. Metastasis from leiomyosarcoma to the eye and periorbital are extremely rare. Conjunctival metastases like in our case are most likely to occur via hematogenous spread as well. Our patient underwent complete surgical excision and was advised to undergo episcleral plaque

brachytherapy for the involvement of base sclera. The status of other distant organ metastasis was unknown as he was non-compliant for further investigations and management. The wound had healed with minimal scarring without any recurrence at 6 weeks and on the last telephonic contact at 9 months he was alive and stable.

In summary, we report a rare case of anterior chest wall leiomyosarcoma metastatic to the conjunctiva. A high index of suspicion regarding possible metastasis should be considered when encountered a solitary conjunctival nodule in a patient with known primary malignancy. Histopathology and immunohistochemistry are essential tools for accurate diagnosis and that help in guiding the optimum management in such rare cases.

Table 1: Review of published cases of conjunctival metastasis (1,9,11-33).

No	Author(s), year	No of cases	Primary malignancy	Conjunctival metastasis site	Treatment
1	Grossniklaus, et al. (1987) [11]	1	Not specified	Not specified	Not specified
2	Jakobiec, et al. (1989) [12]	5	Cutaneous melanoma	Bulbar (2) Fomical (1) Plical-caruncular (1) Palpebral (1)	Excision + treatment of primary malignancy
3	Ortiz, et al. (1995) [13]	1	Uterine cervical carcinoma	Bulbar	Excision + systemic chemotherapy and surgery for primary site
4	Kiratli, et al. (1996) [9]	10	Breast (4) Lung (2) Cutaneous melanoma (2) Larynx (1) Unknown (1)	Bulbar (6) Palpebral (2) Limbal (1) Fornix (1)	EBRT (5) Excision with EBRT (1) Enucleation with EBRT (1) Excision (1) Chemo (1) None (1)
5	Kwapiszewski, et al. (1997) [14]	1	Cutaneous melanoma	Bulbar	Excision + systemic chemotherapy for primary malignancy
6	Shields, et al. (1998) [15]	1	Lung	Bulbar	Enucleation (multiple ocular site involvement)
7	Ziakas, et al. (2000) [16]	1	Cutaneous melanoma	Palpebral	Palliative therapy
8	Alwitry, et al. (2001) [17]	1	Lung	Bulbar	Palliative therapy
9	Tokuyama, et al. (2002) [18]	1	Gastric cancer (signet ring carcinoma)	Bulbar	Excision + Primary site surgery
10	Shields, et al. (2004) [19]	1	Cutaneous melanoma	Bulbar	Excision+ systemic treatment for primary and other metastatic sites
11	Shields, et al. (2004) (1)	13	Breast (5) Lung (3) Cutaneous melanoma (1) Larynx (1) Carcinoid tumor (1) Unknown (1)	Bulbar (13)	Not specified
12	Pompeu, et al. (2005) [20]	1	Renal cell carcinoma	Tarsal	Excision and primary site surgery
13	Skalicky, et al. (2007) [21]	1	Breast	Bulbar	Palliative systemic therapy for primary and other metastatic sites
14	Cano, et al. (2011) [22]	1	Visceral Kaposi sarcoma	Palpebral	Excision
15	Radovanović, et al. (2013) [23]	1	Breast	Bulbar	Excision + palliative radiotherapy + hormonal therapy to primary and other metastasis sites
16	Chung, et al. (2004) [24]	1	Lung (Pleural mesothelioma)	Bulbar	Palliative therapy
17	Park, et al. (2014) [25]	1	Breast	Bulbar	Excision + systemic chemotherapy
18	Chew, et al. (2014) [26]	1	Lung	Palpebral	Excision + radiotherapy to primary and other metastatic sites
19	Sahin, et al. (2014) [27]	1	Lung	Bulbar	Not specified
20	Carey, et al. (2014) [28]	1	Cutaneous melanoma	Bulbar	Excision and systemic chemotherapy for primary site
21	Diltoer, et al. (2015) [29]	1	Breast	Bulbar	Not specified
22	Yadav, et al. (2017) [30]	1	Mucoepidermoid carcinoma of parotid gland	Bulbar	Orbital exenteration
23	Sanchez, et al. (2017) [31]	1	Breast	Bulbar	Palliative therapy
23	Brouwer, et al. (2018) [32]	1	Cutaneous melanoma	Fornix	Excision + systemic chemotherapy
24	Phelps, et al. (2018) [33]	1	Vaginal melanoma	Tarsal	Systemic chemotherapy, Radiation to brain (another metastatic site)
25	Our case, 2020	1	Leiomyosarcoma of Anterior chest wall	Bulbar	Excision + Episcleral plaque brachytherapy (advised)



Acknowledgements

This research has been supported by Desai Brothers Ltd, Pune

Conflict of Interests Statement

The authors have no conflicts of interest to declare.

References

- Shields CL, Demirci H, Karatza E, Shields JA (2004) Clinical survey of 1643 melanocytic and nonmelanocytic conjunctival tumors. *Ophthalmol* 111: 1747-54. <https://doi.org/10.1016/j.ophtha.2004.02.013>
- Nair AG, Kaliki S, Kamal S, Mishra DK, Vemuganti GK (2015) Conjunctival leiomyosarcoma: a report of two cases. *Orbit* 34: 274-8. <https://doi.org/10.3109/01676830.2015.1062034>
- Cajaiba MM, Chojniak MM, Cunha IW (2008) Unusual primary ocular neoplasm in a child: leiomyosarcoma of the ciliary body. *Pediatr Dev Pathol* 11: 479-481. <https://doi.org/10.2350/07-02-0231.1>
- Chaugule SS, Putambekar A, Gavade S, Deshpande R (2019) Primary Orbital Leiomyosarcoma in an Adult Male. *Ophthal Plast Reconstr Surg* 35: e27-9. <https://doi.org/10.1097/iop.0000000000001293>
- Esmali B, Cleary KL, Ho L, Safar S, Prieto VG (2002) Leiomyosarcoma of the esophagus metastatic to the eyelid: a clinicopathologic report. *Ophthal Plast Reconstr Surg* 18: 159-61.
- Vichare NV, Dharmesh V, Rana V (2017) Eyelid metastasis as presenting feature of disseminated leiomyosarcoma—A rare case report. *Saudi J Ophthalmol* 31: 186-9. <https://doi.org/10.1016/j.sjopt.2017.05.008>
- Feinstein E, Kaliki S, Shields CL, Ehya H, Shields JA (2014) Choroidal metastasis from leiomyosarcoma in two cases. *Oman J Ophthalmol* 7: 19-21. <https://doi.org/10.4103/0974-620X.127917>
- Minkovitz JB, Dickersin GR, Dallow RL, Albert DM (1990) Leiomyosarcoma metastatic to the orbit. *Arch Ophthalmol* 108:1525-6. <https://doi.org/10.1001/archophth.1990.01070130027014>
- Kiratli H, Shields CL, Shields JA, DePotter P (1996) Metastatic tumours to the conjunctiva: report of 10 cases. *Br J Ophthalmol* 80: 5-8. <https://doi.org/10.1136/bjo.80.1.5>
- Honavar SG, Manjandavida FP (2015) Tumors of the ocular surface: A review. *Indian J Ophthalmol* 63: 187-203. <https://doi.org/10.4103/0301-4738.156912>
- Grossniklaus HE, Green WR, Luckenbach M, Chan CC (1987) Conjunctival lesions in adults. *Cornea* 6: 78-116. <https://doi.org/10.1097/00003226-198706020-00002>
- Jakobiec FA, Buckman G, Zimmerman LE, La Piana FG, Levine MR, et al. (1989) Metastatic melanoma within and to the conjunctiva. *Ophthalmol* 96: 999-1005. [https://doi.org/10.1016/s0161-6420\(89\)32770-9](https://doi.org/10.1016/s0161-6420(89)32770-9)
- Ortiz JM, Esterman B, Paulson J (1995) Uterine cervical carcinoma metastasis to subconjunctival tissue. *Arch Ophthalmol* 113: 1362-3. <https://doi.org/10.1001/archophth.1995.01100110022012>
- Kwapiszewski BR, Savitt ML (1997) Conjunctival metastasis from a cutaneous melanoma as the initial sign of dissemination. *Am J Ophthalmol* 123: 266-8.
- Shields JA, Shields CL, Eagle RC Jr, Gündüz K, Lin B (1998) Diffuse ocular metastases as an initial sign of metastatic lung cancer. *Ophthalmic Surg Lasers* 29: 598-601.
- Ziakas NG, Eke T, Kendall CH, Goulstine DB (2000) Metastatic cutaneous melanoma to the conjunctiva in an Afro-Caribbean patient. *Eye (Lond)* 14: 667-8. <https://doi.org/10.1038/eye.2000.166>
- Alwitry A, Browning A, Holden R, Joseph A (2001) Conjunctival metastasis: A rare presentation of bronchial adenocarcinoma. *Eye* 15: 677-678. <https://doi.org/10.1038/eye.2001.217>
- Tokuyama J, Kubota T, Otani Y, Egawa T, Wada N, et al. (2002) Rare case of early mucosal gastric cancer presenting with metastasis to the bulbar conjunctiva. *Gastric Cancer* 5: 102-6. <https://doi.org/10.1007/s101200200017>
- Shields JA, Shields CL, Eagle RC Jr, Raber IM (2004) Conjunctival metastasis as initial sign of disseminated cutaneous melanoma. *Ophthalmol* 111: 1933-4. <https://doi.org/10.1016/j.ophtha.2004.04.020>
- Pompeu AC, Arap S, Silva MN, Monteiro DS (2005) Ocular metastasis as first presentation of renal cell carcinoma: report of 2 cases. *Clinics* 60: 75-8. <https://doi.org/10.1590/S1807-59322005000100014>
- Skalicky SE, Hirst LW, Conway RM (2007) Metastatic breast carcinoma presenting as a conjunctival lesion. *Clin Exp Ophthalmol* 35: 767-9. <https://doi.org/10.1111/j.1442-9071.2007.01587.x>
- Cano RG, Cardenas JC (2011) Conjunctival metastasis from Kaposi's sarcoma: A case report. *Diag Cytopathol* 39: 128-31. <https://doi.org/10.1002/dc.21381>
- Radovanović AB, Rasić D, Buta M, Dzodić R (2013) Breast cancer metastasis to the conjunctiva. *Vojnosanit Pregl* 70: 331-4. <https://doi.org/10.2298/vsp120322040b>
- Chung A, Mudhar HS (2015) Conjunctival Metastasis from Pleural Mesothelioma. *Ocular Oncol Pathol* 1: 19-23. <https://doi.org/10.1159/000363768>
- Park YM, Park JH, Lee SU, Lee JS (2014) Metastatic breast cancer presenting as a subconjunctival mass. *J Breast Cancer* 17: 88-90. <https://doi.org/10.4048/jbc.2014.17.1.88>
- Chew R, Potter J, DiMattina A (2014) Conjunctival metastasis as the presenting sign for stage IV lung cancer. *Optom Vis Sci* 91: e38-42. <https://doi.org/10.1097/OPX.0000000000000131>
- Sahin A, Yildirim N, Sahin DG, Basmak H, Acikalin M (2014) Conjunctival metastasis as an initial sign of small cell lung cancer. *Case Rep Med* 2014: 614353. <https://doi.org/10.1155/2014/614353>
- Carey AR, Espana EM, Margo CE (2014) Conjunctival metastasis from a regional cutaneous melanoma. *Can J Ophthalmol* 49: e97-9. <https://doi.org/10.1016/j.cjo.2014.04.014>
- Diltoer E, Ten Tusscher MP (2015) Breast Cancer Metastasis Presenting as Conjunctival Chemosis. *Case Rep Ophthalmol* 6: 439-42. <https://doi.org/10.1159/000442519>
- Yadav R, Battoo AJ, Mir AW, Haji AG (2017) Bulbar conjunctival metastasis from mucoepidermoid carcinoma of parotid—a case report and review of literature. *World J Surg Oncol* 15: 10. <https://doi.org/10.1186/s12957-016-1077-0>
- Sánchez Orgaz M, Gonzalez Pessolani T, Pozo Kreiling JJ, Zamora P, Martí Álvarez C, et al. (2017) Orbital and conjunctival metastasis from lobular breast carcinoma. *Orbit* 36: 197-200. <https://doi.org/10.1080/01676830.2017.1310255>
- Brouwer NJ, Marinkovic M, Jochems A, Kapiteijn EW, van Duinen SG, et al. (2018) Conjunctival Metastasis of a Cutaneous Melanoma. *Ocul Oncol Pathol* 4: 107-111. <https://doi.org/10.1159/000479114>
- Phelps PO, Farber MJ, Meyer DR (2018) Pigmented Conjunctival Lesions as Presenting Signs of Vaginal Melanoma. *Ocul Oncol Pathol* 4: 79-81. <https://doi.org/10.1159/000478278>
- Toro JR, Travis LB, Wu HJ, Zhu K, Fletcher CD, et al. (2006) Incidence patterns of soft tissue sarcomas, regardless of primary site, in the surveillance, epidemiology and end results program, 1978-2001: An analysis of 26,758 cases. *Int J Cancer* 119: 2922-30.
- Matsumoto J, Sugiura H, Morikawa T, Kaji M, Kondo S, et al. (2001) A case of primary leiomyosarcoma of the chest wall successfully resected under the video-assisted thoracoscopic approach. *Ann Thorac Cardiovasc Surg* 7:368-70.
- Tsukushi S, Nishida Y, Sugiura H, Nakashima H, Ishiguro N (2009) Soft tissue sarcomas of the chest wall. *J Thorac Oncol* 4: 834-7. <https://doi.org/10.1097/JTO.0b013e3181a97da3>