




CASE REPORT

REVISED Case Report: an unusual orbital tumor [version 2; peer review: 1 approved, 2 approved with reservations]

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Abstract

Introduction

Orbital lipoma is an extremely rare tumor, representing less than 1% of all orbital tumors. We review the literature and describe the presentation, the differential diagnosis and the management of this tumor.




Case report

We report the case of a 63-year-old patient who was referred for a diplopia with recent hemi-cranial headache. Physical examination showed no exophthalmos nor decrease in visual acuity. The patient complained of diplopia on elevation and oculomotricity examination showed limited elevation of the right eye. The Hess Lancaster test was in favor of a limited course of the right inferior rectus muscle. Magnetic resonance imaging revealed a fusiform tissue process in the right inferior rectus muscle with a fatty signal. A complete excision of the tumor was performed by a transconjunctival approach. Cytopathological examination was consistent with a pleomorphic lipoma. The postoperative period was uneventful. The definitive histopathologic diagnosis was a lipoma. The postoperative Magnetic resonance imaging showed the complete disappearance of the lesion. With 3 years of follow up, there is no sign of recurrence or ocular motility trouble.

Conclusion: Lipomas are rare tumors in the orbit. The clinic is variable depending on the size and the site. The clinical diagnosis is difficult to make. Only histology allows the final diagnosis.

Open Peer Review

Approval Status 

	1	2	3
version 2 (revision) 27 Oct 2023			 view
version 1 19 Jun 2023	 view	 view	

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2. **Amira Trigui**, University of Sfax, Sfax, Tunisia
3. **Kirthi Koka**, Sankara Nethralaya Medical Research Foundation, Chennai, India

Any reports and responses or comments on the article can be found at the end of the article.

Keywords

Lipoma, Orbit, Histopathology, MRI, Surgery.

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REVISED Amendments from Version 1

The appearance of pleomorphic lipoma in imaging studies is variable and not pathognomonic for diagnosis, as variation in the ratio of adipose to non-adipose components results in a wide spectrum of imaging features. MRI features such as the hyper-vascularized tissue process and the hypointense T1 and hyperintense T2 character are explained by the presence of a high proportion of non-adipose elements pleomorphic lipoma. The small size of the tumor and its early diagnosis explain the absence of exophthalmos.

In this new version, we have replied to reviewers' comments and made the following changes:

We've added a new reference (7) that also confirms our MRI findings, which show the tumor to be hypointense on T1 and hyperintense on T2.

The expression "transconjunctival" has been replaced by "transconjunctival".

We have replaced the expression "cytopathological" by "histopathological".

This symptom "hemi-cranial headache" has been added in the case report section.

We've added a new expression "General examination revealed no other lipoma locations"

We've added a new expression "bilateral diplopia"

Any further responses from the reviewers can be found at the end of the article

Introduction

Lipomas are benign tumors, rare in the orbit, representing less than 1% of all orbital tumors. They pose a differential diagnosis with a variety of other expansive orbital masses.¹ We report a new case, review the literature and discuss the clinicopathological and radiological features, the differential diagnosis and the management of this entity.

Case report

A 63-year-old unemployed Tunisian woman, with no previous personal or family pathological history, presented with a diplopia with recent hemicranial headache evolving for two weeks. Physical examination showed no exophthalmia and no decrease in visual acuity. Furthermore, it revealed bilateral diplopia on elevation. Oculomotricity examination showed limited elevation of the right eye, which was confirmed by the Hess Lancaster test that revealed a limited course of the right inferior rectus muscle. General examination revealed no other lipoma locations. Magnetic resonance imaging (MRI) showed a fusiform and hyper-vascularized tissue process located in the right inferior rectus with fatty signal. The tumor was hyperintense on spin-echo T2-weighted images (Figure 1) and hypointense on spin-echo T1-weighted images (Figure 2).



Figure 1. Sagittal section of orbital MRI showing a mass with high signal on a T2 weighted image (red arrow).

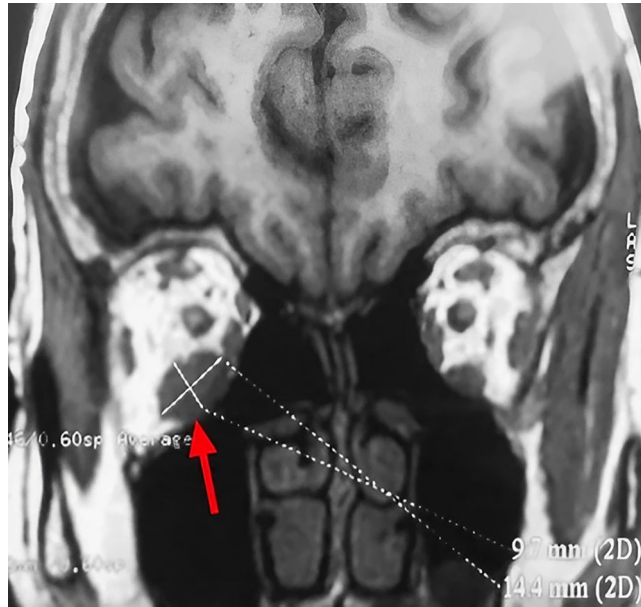


Figure 2. Coronal section of orbital MRI showing a mass with low signal on a T1 weighted image (red arrow).



Figure 3. Macroscopic appearance of the specimen.

These findings suggested various diagnosis; lipoma, inflammatory process, lymphoma and malignant tumor.

We performed a right inferior transconjunctival orbitotomy and excisional biopsy under general anesthesia. Intraoperatively, we discovered an encapsulated mass of fatty tissue, thus complete excision was made. No adhesions or involvement of adjacent structures occurred. The specimen was well circumscribed and slightly firmer than normal adipose tissue, with a yellow surface (Figure 3).

Histologic examination was consistent with a pleomorphic lipoma. The postoperative period was uneventful. Immediately after the operation, the patient reports the resolution of his diplopia. Postoperative MRI images demonstrated the complete resolution of the tumor (Figure 4). With 3 years of follow up, there is no sign of recurrence or ocular motility impairment.

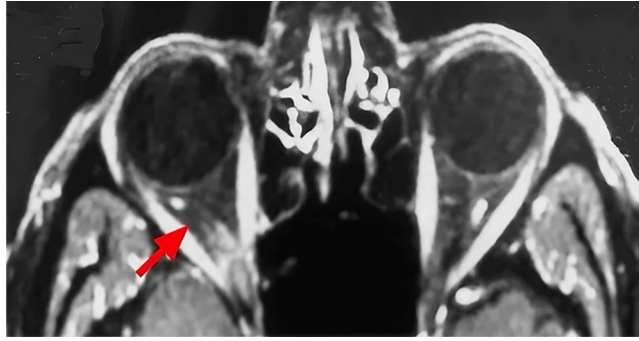


Figure 4. Postoperative axial MRI image demonstrating complete tumor resolution (red arrow).

Discussion

Orbital lipoma is the most common mesenchymal soft tissue tumor. However, it is rarely found in the orbit despite the presence of abundant adipose tissue in the intraorbital space.^{2,3} A review of the largest series of orbital tumors revealed a very low incidence of lipomas.⁴ Shields *et al.* reported only two cases of lipomas in a review of 1264 cases of orbital tumors, indicating the rarity of this entity.⁵ On physical examination, the diagnosis is often difficult to suggest. These tumors are often asymptomatic.

However, they can cause severe morbidity by causing progressive and painless exophthalmos, which is occasionally coupled with diplopia or ocular motility defects⁶ such as was observed in our patient.

Orbital lipoma exceptionally leads to a compressive neuropathy responsible for a significant decrease in visual acuity, an alteration of the afferent photomotor reflex and the visual field constriction.¹ Imaging based on computed tomography (CT) scanning and MRI is essentially useful in ascertaining determining the exact seat, size and relationship to the orbit content. The fatty signal is characteristic on CT sequences. Furthermore, as was found in our patient, the tumor is hypointense on spin-echo T1-weighted images and hyperintense on spin-echo T2-weighted images.⁷

Histology is essential for definitive diagnosis of pleomorphic lipoma. An important histologic criterion is the presence of a mixture of fat cells, pleomorphic cells and in particular floret-like multinucleated giant cells embedded in a myxoid stroma.⁸ That concurred with the histological result of our case. Differential diagnosis of this tumor became more important because the number of reports about some other tumors of similar morphology, are increasing. Pleomorphic lipoma may be confused with lipomatous hemangiopericytoma, myofibroblastoma or even malignant tumors such as rhabdomyosarcoma, myxoid malignant fibrous histiocytoma and liposarcoma.^{9,10} Surgical excision of an orbital lipoma is not only recommended for symptomatic cases such as our patient's clinical presentation but also to exclude malignancy.¹¹ In addition, as was noted in our patient, the long-term outcome after surgery is considered excellent.¹²

This case highlights the importance of orbital imaging in the context of diplopia without obvious cause to rule out an intraorbital lipoma. Nevertheless, this association remains rare and requires further documentation of cases.

Conclusion

Lipomas are benign soft tissue tumors, rarely located in the orbit. The clinical presentation is variable depends on the size and the intraorbital site. The histology makes the definitive diagnosis and may precisely identify the variant.

Consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Data availability

Underlying data

All data underlying the results are available as part of the article and no additional source data are required.

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Current Peer Review Status: ? ✓ ?

Version 2

Reviewer Report 08 May 2024

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Kirthi Koka

Orbit, Oculoplasty, Reconstructive and Aesthetic Services, Sankara Nethralaya Medical Research Foundation, Chennai, Tamil Nadu, India

The authors have presented an interesting case of orbital lipoma.

The authors have reviewed literature but there is no mention of the different variants of lipomas and how one can differentiate between these.

The authors have added imaging features on MRI which is suggestive of hypointense lesion on T1 weighted images.

But as per most literature, Lipomas being predominantly made up of fat, are hyperintense on T1 weighted image (as also documented in one of the references cited in the article - reference no 12). This does not correlate with the imaging findings and descriptions. Kindly clarify the same

Also as per the imaging findings the lesion seems to be infiltrating into the inferior rectus muscle. The authors need to elaborate the surgical findings on table, if muscle fibres were involved or if the lesion was a well encapsulated, separate lesion that could be easily dissected and removed with no damage to the muscle fibres.

In discussion the authors mention that imaging helps confirm the exact seat. Kindly elaborate what they mean by seat.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Partly

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Orbit and Oculoplasty

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Version 1

Reviewer Report 16 October 2023

<https://doi.org/10.5256/f1000research.142784.r193958>

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Amira Trigui

Department of Ophthalmology, University of Sfax, Sfax, Tunisia

A 63-year-old man presented with diplopia for two weeks. MRI of the brain and orbit revealed a process in the orbit with a fatty signal. Excisional biopsy of the mass resolved the diplopia, and histology revealed a pleomorphic lipoma. This report may help clinicians in the future to be alert to the possibility of the presence of an orbital tumor despite the absence of exophthalmos, which is a common finding in this setting. In this case, the clinical observation is detailed and the MRI images are of excellent quality, but a few points need to be clarified:

- Did the patient's general examination reveal obesity, or other lipoma locations?
- Did the authors perform orbital ultrasound before MRI imaging?
- Authors should specify whether diplopia is monocular or binocular.
- It would also be interesting if the authors could explain the absence of exophthalmos in this case.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment

given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.**Reviewer Expertise:** Ophthalmology**I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.**

Reviewer Report 04 August 2023

<https://doi.org/10.5256/f1000research.142784.r190823>

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**Mehdi Hasnaoui**

Department of Otolaryngology-Head and Neck Surgery, Tahar Sfar Hospital, Mahdia, Tunisia

Editorial Note from F1000Research – 14 August 2023: A COI statement has been added detailing a shared affiliation between the authors and reviewer, which was not declared at the time of the publishing of this report. The COI statement is below.

This article is well written. It is easy to read. The figures are demonstrative. The authors report a rare case of an orbital lipoma revealed by isolated diplopia. The MRI was not very suggestive of a lipoma. The authors insisted on the interest of MRI in the context of diplopia. They discussed the clinicopathologic and radiological characteristics and the differential diagnosis of this entity.

I have some minor comments:

Abstract :

- The authors state: *"We report the case of a 63-year-old patient who was referred for a diplopia with recent **hemi-cranial headache**". This symptom "**hemi-cranial headache**" was not mentioned and written in the case report section.*
- The authors state: *"**Cytopathological** examination was consistent with a pleomorphic lipoma.*

The postoperative period was uneventful. The definitive histopathologic diagnosis was a lipoma." Did the authors do a cytopathological examination and then a definitive histological examination? This examination "cytopathological" was not mentioned and written in the case report section.

- Line 13 : change "trasncunjonctival" to "transcunjonctival"

Case report :

- The authors state: "*MRI showed a spindle-shaped and hyper-vascularized tissue process*". How to explain this hypervascularization?
- Do the authors have postgadolinium fat-saturated axial T1 images? Lipomas are classically hypointense after fat suppression.

Discussion :

- In this case, the tumor was hyperintense on spin-echo T1-weighted images. This is not usual in lipomas and should be discussed.
- The authors state: "Furthermore, as was found in our patient, the tumor is hypointense on spin-echo T1-weighted images and hyperintense on spin-echo T2-weighted images.¹" However, in the literature and in the cited reference number 1, most lipomas are generally hyperintense on T1-weighted imaging.

I believe the paper has academic merit, but minor revision is needed.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Partly

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: This reviewer is affiliated at Tahar Sfar University Hospital, which is the same as the authors of this article. This was not declared as a conflict of interest at the time of publication of this report but this reviewer can confirm that this did not influence the impartiality of this report.

Reviewer Expertise: Oncology, sinus and nasal cavity surgery, facial bone imaging

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 04 Aug 2023

Anis Mahmoud

I would like to thank the reviewer for his interest in this work and will answer his queries in this report:

The MRI appearance of pleomorphic lipomas is variable and not pathognomonic for diagnosis, as the variation in the ratio of adipose to non-adipose components results in a wide spectrum of imaging features. MRI features such as hyper-vascularized tissue process and hypointense T1 and hyperintense T2 character can be explained by the presence of a high proportion of non-adipose elements in pleomorphic lipomas.

We have added a new reference (7) that also confirms our MRI results, which show the tumor to be hypointense in T1 and hyperintense in T2.

The expression "trasncunjonctival" has been replaced by "transconjunctival".

Cytopathological" has been replaced by "histopathological".

The symptom "hemi-cranial headache" has been added to the "case report" section.

Competing Interests: No competing interests were disclosed.

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