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Unilateral peri-orbital oedema and mechanical ptosis: an unusual case presentation of rosacea

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Title Page

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Unilateral peri-orbital oedema and mechanical ptosis: an unusual case presentation of rosacea

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Abstract

Introduction: Rosacea is an inflammatory skin condition that can present with varied ophthalmic manifestations. It is often overlooked by clinicians especially when unilateral in presentation leading to diagnostic delay and a resultant psychosocial impact. We aim to present a unique case of ocular rosacea, highlighting the difficulty in therapeutic challenges and diagnoses in such rare cases.

Case presentation: A 64-year-old Caucasian man presented with a nine-month history of persistent painless swelling of the right upper eyelid and secondary ptosis. His ophthalmic examination, serology and MRI were otherwise normal besides mild meibomian gland dysfunction. Punch biopsy results were inconsistent and initially led to a misdiagnosis of benign squamous papillomata and later, a differential diagnosis of dermatomyositis. He was trialled on appropriate management for these conditions without any benefit. Repeat histopathology was suggestive of rosacea and given the persistence of symptoms despite multiple treatments, he was successfully managed with a right upper lid debulking biopsy transcutaneous blepharoplasty. Histopathological analysis of the debulking biopsy confirmed the diagnosis of rosacea, with additional features indicative of lymphoedema. Upon follow up, there was resolution of lid swelling.

Conclusion: Due to the nonspecific nature of isolated ocular rosacea presentations, it can be easily misdiagnosed and therefore, should always be considered as a differential diagnosis in persistent peri-orbital oedema. It can additionally pose significant therapeutic challenges for ophthalmologists, underscoring the importance of

improving our understanding of ocular rosacea. Further, we have shown the effectiveness of surgical debulking in its management.

Keywords: eyelid oedema, eyelid ptosis, rosacea, debulking blepharoplasty, interface dermatitis

Introduction

Rosacea is a chronic inflammatory disorder that commonly affects individuals of all skin types. Its varied presentation can lead to ocular manifestations that are overlooked by ophthalmologists, which may result in a delay in diagnosis, subsequently leading to significant psychosocial impacts on patients and potential vision loss if left untreated. Here, we present a case of ocular rosacea with unilateral right upper-eyelid oedema resulting in secondary ptosis, reported in accordance with the CARE Checklist (see online supplementary material).

Case report

A 64-year-old Caucasian man presented with a 9-month history of persistent painless swelling of the right upper eyelid with a sensation of grittiness, both worse in the morning and diminishing throughout the day. He had a past medical history of gout and no previously known ocular or dermatological conditions. His only regular medication was allopurinol 100mg once a day. There was no significant family history.

External examination revealed oedema of the right upper eyelid, particularly laterally and extending to lateral canthal area (figure 1a). The oedema was soft with no evidence of pitting and no tenderness on palpation. There was no erythema nor signs of inflammation, however, mechanical ptosis was evident. The left eye exhibited a normal MRD1 of 5mm whilst the right eye measured 0mm. MRD2 was normal bilaterally, measuring 6mm in the left eye and 5mm in the right. Function of the orbicularis, levator and frontalis muscles were intact bilaterally, with both eyes portraying full ocular motility. There were no signs of Bell's palsy, Marcus Gunn Jaw Winking Syndrome nor fatiguability.

Visual acuity was 6/7.5 bilaterally. Ocular examination identified bilateral mild meibomian gland dysfunction and Terrien's margin degeneration. The eyes were otherwise white and quiet with normal fundoscopic appearances. Lid eversion identified no papillae, follicles or other abnormality. Pupils were equal and reactive. Intraocular pressures were normal and confrontational visual field testing revealed no deficit in either eye. Refraction was +1.5/-0.5x65 OD and +1.5/-0.25x30 OS. There was no evidence of pre-auricular or submandibular lymphadenopathy.

The following laboratory studies were normal: urea & electrolytes, thyroid function tests, HBA1c, C3, C4, C1 Esterase Inhibitor and Functional C1 Esterase Inhibitor. Magnetic resonance imaging of the head and orbits confirmed pre-septal swelling of the right eyelids and no evidence of cranial or orbital pathology (figure 2).

A 2mm punch biopsy was taken, leading to an initial diagnosis of benign squamous papillomata of the right upper lid. The patient was trialled on oral Aciclovir 400mg three times daily and right eye Aciclovir ointment at night for three months. Upon follow up, there was no change in clinical appearances and other examination findings remained unchanged. Given the concurrent meibomian gland dysfunction, the patient was trialled on doxycycline 100mg once a day, 0.2% sodium hyaluronate preservative free twice a day (Xialin® Plus), Hylo Night® ointment once at night and daily lid hygiene for 6 weeks. Cool compresses to both eyelids and avoidance of direct sunlight exposure to the skin was additionally recommended.

On review three months later, there was a worsening of oedema and secondary ptosis. This led to further considerations of problems with eyelid lymphatic drainage and widened the differential to include lymphoedema, blepharochalasis and possible malignancy. Therefore, the patient underwent further multiple punch biopsies which were sent for both microbiological and histopathological analysis. His microbiology results revealed no organisms on gram staining and on culture, *Cutibacterium acnes*, but only on enrichment. His histopathology results revealed epidermal spongiosis, patchy vacuolar interface and subepithelial mixed inflammatory infiltrate of lymphocytes, histiocytes and plasma cells mainly in periadenexal and perivascular distribution, features suggestive of interface dermatitis. On immunohistochemistry, the lymphoid component was predominantly CD3 positive T cells, and these were a mixture of CD4 and CD8 positive lymphocytes. There was no evidence of dysplasia or malignancy.

The case was discussed at a joint Dermatology & Ophthalmology Multidisciplinary Team (DOMDT) Meeting. Given the clinical findings and the absence of features suggestive of lymphoma or malignancy, a diagnosis of dermatomyositis was considered by consultant dermatologists on review of his clinical photographs. Dermatological examination, however, did not show any other features of dermatomyositis, his chest X-Ray was normal, and urinalysis was performed to rule out nephrotic syndrome, which showed microhaematuria without significant protein. A diagnosis of probable lymphoedematous rosacea was made and dermatology recommended doxycycline 50mg once daily for a month and Acitretin for mitigating ocular swelling, with advice of sitting up whilst sleeping and daily eyelid massages.

On further follow up three months later, the patient reported improvement in peri-orbital swelling with a significant subjective change. This improvement occurred within the first three weeks of starting Acitretin with a lack of sustained improvement after this time. However, Acitretin also resulted in adverse effects including muscle aches and elevated triglycerides resulting in the joint decision to discontinue it. The patient's symptoms then worsened with increased oedema and significant eyelash ptosis (figure 1b), which threatened the integrity of his ocular surface. He was again discussed at a joint DOMDT meeting, this time with a consultant pathologist present. A review of the patient's previous pathology slides in the meeting confirmed features highly suggestive of rosacea. In view of the patients worsening symptoms, surgical options were discussed.

A right upper lid debulking biopsy and transcutaneous blepharoplasty was performed, three years after initial presentation. All excised tissue samples of eyelid skin with subcutaneous tissue muscle were sent for histological analysis. This revealed minimal focal basal layer vacuolar degeneration of epidermis, dermal oedema and prominent perifollicular and perivascular inflammatory cell infiltrates. The inflammatory cells were a mixture of small lymphocytes, plasma cells and histiocytes (figure 3a). There was no presence of granulomas, basement membrane thickening, follicular plugging or evidence of vasculitis, dysplasia and malignancy. On higher magnification, peri-infundibular infiltrate of lymphocytes and histiocytes with focal follicular spongiosis (visible on the left side of follicle) were spotted, accompanied by exocytosis of inflammatory cells into follicular epithelium (figure 3b). Telangiectasia was also present. Elongated, empty ectatic papillary dermal vessels were observed (figure 3c), and highlighted with brown D2-40 immunostaining confirming lymphangiectasia, supporting the presence of lymphoedema (figure 3d). The histological features were entirely consistent with right upper lid rosacea, with evidence of lymphangiectasia and lymphoedema. At follow up, there was significant improvement in clinical findings with resolution of majority of the patient's lid swelling (figure 1c). On subsequent review, whilst there remained complete resolution of the ptosis, a shallow area of chronic oedema was noted (arrow, figure 1c). It was, therefore, decided to trial a 20mg (0.5mls) subcutaneous injection of triamcinolone which cleared the remaining oedema completely. A summary of all treatment intervention and outcomes is shown in table 1 and a detailed timeline of the patient's clinical journey is shown in table 2.

Discussion

Rosacea is a common condition that affects 5.5% of the global population, with up to 58%-72% of patients developing ocular surface and or lid margin involvement either before or after cutaneous disease [1-3]. Presentations of rosacea varies depending on individuals; however, symptoms and signs can include frequent episodes of flushing, persistent erythema, telangiectasia, papules, pustules and rhinophyma [4]. Isolated eyelid lymphoedema not involving the face, as in our case, is a complication rarely mentioned in literature. Upper facial

lymphoedema of rosacea involving the forehead, eyelids and cheeks has been referred to as 'solid facial oedema of rosacea', 'Morbihan's disease' or 'persistent facial oedema of rosacea'. Isolated unilateral upper eyelid involvement, without involvement anywhere else, appears to be even more uncommon, with only one published report refractory to medical therapy and others reports describing swelling in the lower eyelids or involvement of other areas of the face [2, 3, 5–9]. To the knowledge of the authors our case is the first reported case of isolated unilateral upper lid rosacea that shows a patient journey through several stages of clinical management and successfully managed by debulking upper eyelid surgery, when refractive to medical therapy.

Due to the variety of presentations of ocular rosacea, differential diagnoses are wide and may include several groups of diseases. Isolated ocular rosacea may present similarly to blepharitis, allergic, bacterial or viral conjunctivitis, allergic dermatitis, dermatomyositis, angioedema, inflammation and malignancies posing further challenges in reaching a diagnosis of ocular rosacea [3, 10].

The pathophysiology of ocular rosacea remains unknown. Similar to hypothetical models of cutaneous rosacea, it has been reported that dysregulation of neural control of vascular flow may contribute to enlargement of ocular surface blood vessels, related to elevated levels of transient receptor potential channels and vascular endothelial growth factors found in rosacea patients [11, 12]. However, it is not elucidated whether the mechanism is present as a cause or consequence of inflammatory changes within pathology progression. It is also advocated that dysregulation of the innate and adaptive immune systems may induce the release of proinflammatory markers and antimicrobial peptide levels, such as elevated levels of LL-37, which then synergistically maintains chronic inflammation alongside abnormal vascular control [12, 13]. Meibomian glands are also a recognised major component of rosacea pathophysiology, with its dysfunction prevalent in most ocular rosacea cases [14]. Other theories postulate elastin damage as the main cause of vascular dysregulation, leading to reduced vessel wall integrity, consequently resulting in the transudation of fluids and oedema [12, 15]. It has also been mentioned that the same mechanism results in fibrosis and permanent obstruction of deep dermal lymphatic vessels, resulting in fluid accumulation and lymphoedema [15].

There is limited literature regarding histopathology findings of ocular rosacea, with some stating that there is no histological pattern unique to rosacea, supporting the multi-factorial origin of the disease [16]. Conjunctival epithelial findings include infiltration of inflammatory cells, mainly of T helper/ inducer (CD4) cells, phagocytic cells and antigen presenting CD14 cells [17]. Other skin biopsy findings include solar elastosis and dilated vascular channels with a predominantly lymphohistiocytic inflammatory infiltrate, sometimes with granulomas [16, 18]. Our case demonstrated significant findings of peri-infundibular and perivascular lymphohistiocytic inflammatory infiltrates, with predominantly CD3-positive T cells. We identified no granulomas, but follicular epithelial and subepithelial inflammatory infiltrates, especially in periadenexal and perivascular distribution, are likely contributors to oedema in this case.

Similarly, histological features specific to lymphoedema in rosacea remain poorly defined, with literature reporting lymphocytoclasis in swollen lymphatic channels and lymphangiectasia as potential indicators [2, 3]. Whilst our case did not exhibit lymphocytoclasis, the presence of multiple empty and elongated lymphatic channels proven by D2-40 immunostaining (lymphangiectasia) strongly supports a possible lymphoedema manifestation.

There are multiple treatment options for ocular rosacea, but no established treatment guidelines nor consensus regarding the management of ocular rosacea, including the specific approach for rosacea-related ocular lymphoedema. Similar cases have reported the use of systemic corticosteroids, though most cases showed no effect. Other treatment options include antihistamines, isotretinoin and oral antibiotics, such as clofazimine, topical metronidazole and oral macrolide. Some reports also mention the use of thalidomide, with isotretinoin considered as a first line treatment despite the lack of consistent efficacy [7-9]. In our case, the patient failed to significantly respond conservative medical measures, with worsening symptoms necessitating surgical debulking. Interestingly, surgical outcomes have ranged from recurrence to complete resolution, highlighting the variability in treatment response [3, 8, 19]. Successful cases of surgical debulking have been reported on few occasions, including peri-orbital oedema due to Morbihan syndrome [3, 19, 20].

In conclusion, unilateral upper eyelid lymphoedema in rosacea is a rare presentation. Due to the nonspecific nature of the condition, it can present significant diagnostic and therapeutic challenges for ophthalmologists. This underscores the importance of highlighting rare cases such as this.

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Statements and Declarations

Statement of Ethics

Ethical approval is not required for this study in accordance with local guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images, in line with Aneurin Bevan University Health Board guidelines.

Conflict of Interest: No Competing Interests

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Author Contributions:

SWRL – data collection, compilation and analysis, primarily wrote the manuscript
NC – reviewed data analysis and co-drafted the manuscript
LI – offered expert histopathological guidance on the case and reviewed the manuscript
NS – offered expert dermatological guidance on the case and reviewed manuscript
EO – study design, served as the primary physician managing the patient case, acquired and interpreted clinical data and reviewed the manuscript
All authors approved the final manuscript

Availability of data and materials: All data generated and analysed for the case report are included in this article. Any further enquiries regarding the case can be directed to the corresponding author.

Figure Legends

Figure 1

Clinical appearances **(A)** At initial presentation, there is oedema and secondary ptosis of the right upper eyelid particularly laterally and extending below right lower lid. At follow up after an incidental initial diagnosis of benign squamous papillomata – there was no change in clinical appearance. **(B**) Subsequent ophthalmic follow up after dermatology review and treatment. Worsening oedema, mechanical ptosis and significant lash ptosis. **(C)** Complete resolution of the mechanical ptosis and majority of the oedema after a debulking blepharoplasty.

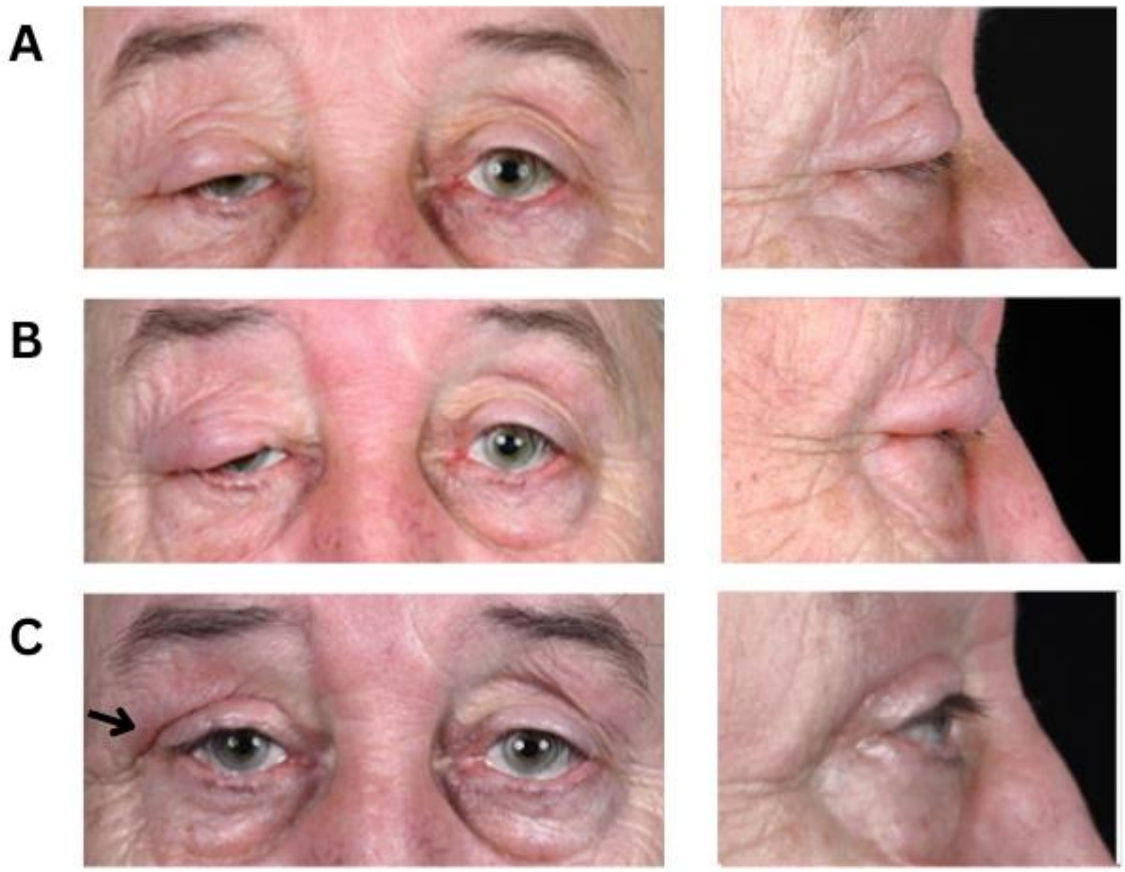
Figure 2

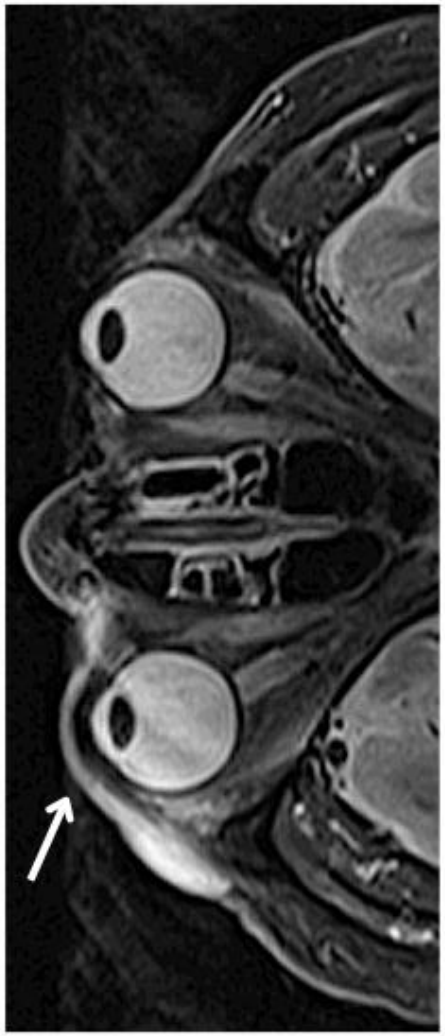
Magnetic Resonance imaging of head and orbits showing pre-septal swelling of right eyelids

Figure 3

Histopathology slides **(A)** Low power view, prominent perifollicular and perivascular lymphohistiocytic infiltrates present. **(B)** Higher magnification, peri-infundibular infiltrate of lymphocytes and histiocytes seen with focal follicular spongiosis and exocytosis of inflammatory cells into follicular epithelium. **(C)** Presence of ectatic

papillary dermal vessels. **(D)** D2-40 immunostain, brown, confirming the dilated vessels represent lymphatic channels-lymphangiectasia





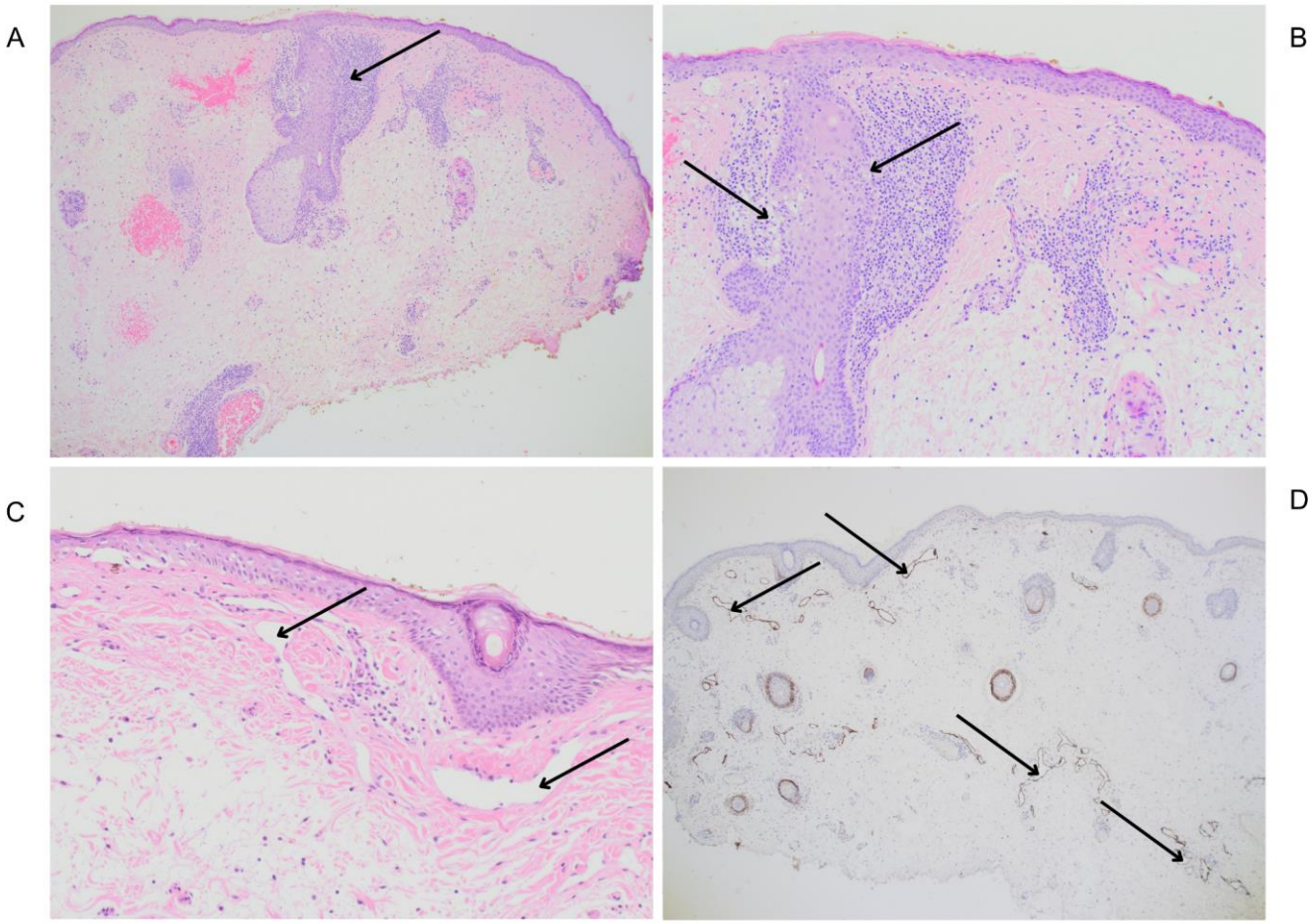


Table 1. Treatment outcomes for each treatment trialled in chronological order

Time (number of follow up)	Treatment	Treatment duration	Treatment outcomes
April 2023 (1st)	Oral aciclovir 300mg TDS	3 months	• No change in clinical appearance nor examination findings • Treatment stopped
August 2023 (2nd)	Oral doxycycline 100mg OD Topical 0.2% sodium hyaluronate preservative free BD Topical Hylo Night ointment nocte Avoidance of direct sunlight exposure Daily lid hygiene	6 weeks	• No improvement. Clinically there was worsening of oedema and secondary ptosis on follow up review • Treatment stopped
April 2024 (3rd)	Oral doxycycline 50mg OD Acitretin	1 month	• Initial improvement but not sustained after 3 weeks of initiation • Adverse drug effect - significant aches in arms and legs reported by patient • Treatment stopped
September 2024	Right upper lid debulking biopsy Transcutaneous blepharoplasty	N/A	• Complete resolution of ptosis • Resolution of significant oedema
June 2025	Subcutaneous 20mg (0.5ml) Triamcinolone Injection	N/A	• Complete resolution of oedema

Table 2. Detailed timeline of the patient's clinical journey

Dates	Events
Sept 2021	• Initial presentation to GP resulting in Ophthalmology referral
Jan 2022	• First Ophthalmology assessment including external examination, visual function testing and laboratory studies completed.
May 2022	• Contrast MRI scans of Head and Orbits – evident visible pre-septal swelling of right eyelid, otherwise normal • Angioedema assessment including complement studies
Oct 2022	• 2mm punch biopsy of right upper lid taken which revealed an incidental diagnosis of benign squamous papillomata of right upper lid
Jan 2023	• Trialled on oral aciclovir 300mg three times a day + right eye aciclovir ointment at night for 3 months
Apr 2023	• No change in clinical appearance nor examination findings • Concurrent meibomian gland dysfunction managed with prescribed doxycycline 100mg once a day, 0.2% sodium hyaluronate preservative free twice a day, Hylo Night ointment once at night, cool compresses, avoidance of direct sunlight exposure and daily lid hygiene for 6 weeks
Aug 2023	• Worsening of oedema and secondary ptosis • Repeat right upper lid biopsy - sent for repeat histopathology, microbiology and immunohistochemistry evaluation. Histological features suggestive of Interface Dermatitis and Lymphoma excluded.
Nov 2023	• Case Discussion at Dermatology-Ophthalmology Multi-Disciplinary (DOMDT) Meeting • Differential diagnosis of dermatomyositis by the dermatologists on review of the patients photographs. We were advised to refer the patient to the dermatology clinic for assessment
Jan 2024	• Dermatology review including full examination, chest X-ray and urinalysis • Formal diagnosis of lymphoedematous rosacea • Started on oral doxycycline 50mg once a day for 1 month, acitretin for ocular swelling, eyelid massages and positional changes whilst sleeping recommended
Apr 2024	• Initial improvement in oedema, however, not sustained after 3 weeks of initiation • Reported significant adverse effects from acitretin including aches in arms and legs
Jul 2024	• Re-discussion in DOMDT Meeting, with another consultant Pathologist at the meeting. • Histology re-examination confirmed features of rosacea so surgical options were considered
Sep 2024	• Right upper lid biopsy and transcutaneous debulking biopsy and blepharoplasty completed • Excised tissues sent to histopathology - this confirmed again the diagnosis of rosacea

Apr 2025	• Sustained improvement with no recurrence of significant oedema • Very mild area of shallow chronic oedema – underwent 0.5mls (20mg) subcutaneous injection of triamcinolone
Jun 2025	• Complete resolution of oedema