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Holding back the tears: is there a role for marsupialisation?

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ABSTRACT

Objective Medial eyelid tumours may result in the loss of the proximal lacrimal system during staged excision and delayed reconstruction, to achieve tumour margin clearance. The remnant canaliculus was marsupialised during reconstruction. The aim was to understand how many patients experienced symptomatic epiphora as a consequence of this.

Methods and analysis A retrospective study including patients over a 15-year period with medial eyelid tumours, where the proximal lacrimal system was sacrificed to achieve tumour margin clearance. Included were all who had marsupialisation of the remnant distal stump as part of their delayed reconstruction. All who had pre-existing epiphora were excluded. The primary objective was the rate of epiphora following the procedure. A systematic literature review of postoperative epiphora occurring in patients with lid tumours requiring lacrimal system injury/sacrifice during tumour excision.

Results There were 22 eyes (22 patients). All were basal cell carcinomas except for 1 (4.5%) tarsal conjunctival squamous cell carcinoma. All cases involved the lower lid. There were two (9.1%) patients who developed epiphora. One patient underwent a superior three-snip punctoplasty, botulinum toxin to the lacrimal gland and conjunctivodacryocystorhinostomy with Lester Jones tube insertion. The other patient was not overly troubled and did not require further treatment. The literature review showed the median postoperative rate of epiphora in these patients was 12.5% (range 0%–100%).

Conclusion Marsupialisation of the remnant canaliculus during delayed reconstruction is a straightforward and effective surgical option, which may help prevent postreconstruction epiphora when the proximal lacrimal system is sacrificed for tumour margin clearance.

Trial registration number 10391.

WHAT IS ALREADY KNOWN ABOUT THIS TOPIC

⇒ Basal cell carcinomas are the most common eyelid tumours in the Caucasian population, and the second most common location is the medial canthal area.

⇒ Tumour clearance in this area poses challenges due to the complex anatomy of the medial canthus and its proximity to the nasolacrimal apparatus, and failure to rehabilitate the nasolacrimal apparatus during eyelid reconstruction may result in potentially disabling epiphora.

WHAT THIS STUDY ADDS

⇒ The literature was reviewed to identify only those cases with lacrimal system injury/sacrifice occurring during tumour resection, and the rate of epiphora in those patients. The median was 12.5% (range 0%–100%).

⇒ In this study, the rate of postoperative epiphora following marsupialisation of a remnant canaliculus in this context was 9.1% (2/22), but only 4.5% (1/22) required further treatment.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ Marsupialisation of the remnant canaliculus during delayed eyelid reconstruction is a straightforward, effective and cost-efficient surgical option which may help prevent postoperative epiphora when the proximal lacrimal system is sacrificed for tumour margin clearance.

INTRODUCTION

Periocular malignancies are estimated to occur in 5%–10% of all skin cancers.[1] Of newly diagnosed malignancies, skin cancers comprise about one-third.[2] Basal cell carcinomas are the most common malignant eyelid tumours in the Caucasian population, estimated to occur in 85%–95% of the population.[1 3] The second most common location for their occurrence is in the medial canthal area.[1] Reconstruction of the medial canthal region following excision of such tumours poses specific challenges, owing to both the complexity of the anatomy in the medial canthus and its importance in eyelid function and vision.[4] Normal anatomical positioning of the eyelids and lid margins on the ocular surface and intact physiological pump mechanisms are required for effective tear clearance from the ocular surface into lacrimal outflow.[5] Furthermore, the nasolacrimal apparatus requires consideration during reconstruction, as failure to repair defects to the nasolacrimal apparatus may result in potentially disabling epiphora.[6] Loss of some or all of the nasolacrimal apparatus may be required in order to achieve tumour margin clearance.

Various methods for restoring proximal lacrimal system patency have been described, irrespective of whether the aetiology was due

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to trauma, tumour resection, or punctal or canalicular stenosis or obstruction. For punctal stenosis/obstruction, treatment methods include perforated punctal plugs, punctal snip procedures, punctal marsupialisation or recreation of a punctal opening.[7 8] These may be combined with recanalising procedures for the canaliculus. For restoring canalicular patency, these methods include balloon canaliculoplasty, canalicular trephine, and monocanalicular or bicanalicular stenting.[9] Bypassing the canaliculus and lacrimal sac altogether with a conjunctivodacryocystorhinostomy and Lester Jones tube placement remains the gold standard when there is extensive canalicular obstruction, congenital agenesis, failed previous canalicular surgery or functional epiphora.[9] When part of the proximal lacrimal system is compromised due to tumour or trauma, there also remains the option of leaving the remnant lacrimal system alone without reconstruction and only reconstructing the lid defect,[10] or leaving the lid defect to heal by secondary intention.[11]

Lacrimal system reconstruction following medial canthal tumour resection is not well studied. The majority are case series or a small subset of patients in larger retrospective cohort studies that examine either a specific reconstructive method to the skin and medial canthal area or lacrimal system obstruction due to a variety of causes.[10–16] Marsupialisation of the remnant canaliculus is a relatively straightforward procedure. It can be easily incorporated into the reconstruction of the eyelid following tumour resection. If successful, it would obviate the need for further rehabilitative surgery, which may be more complex and involve long-term upkeep, such as in the case of using Lester Jones tubes for lacrimal bypass. This study aimed to examine the outcomes in patients who underwent marsupialisation of a remnant canaliculus as part of eyelid reconstruction, when the proximal lacrimal system was lost during tumour excision to achieve tumour margin clearance. The primary outcome was the number of patients who had epiphora.

MATERIALS AND METHODS

This was a retrospective study covering a period of 15 years (September 2004–January 2020) at a British tertiary centre. The study was registered with the Clinical Effectiveness Unit of Sheffield Teaching Hospitals as a service evaluation project, in line with guidance in the National Health Service (NHS). All data retrieved on individual patients were handled in a secure and confidential manner and stored in password protected NHS computers to maintain patient privacy. The data were anonymised prior to analyses and reporting.

Patients with medial eyelid tumours were identified from the surgeon's logbook (JHYT). In this unit, most of the eyelid cancer reconstructions are done by this surgeon (JHYT) or by an ophthalmology trainee or oculoplastics fellow under her direct supervision. All patients underwent excision of their eyelid tumour with delayed reconstruction once tumour margin clearance was confirmed. It is the standard practice of this surgeon (JHYT) to marsupialise any remnant canaliculi, if the proximal lacrimal system is sacrificed to achieve tumour margin clearance. All patients with marsupialisation of a remnant canalicular stump following sacrifice of the proximal lacrimal system to achieve tumour margin clearance were included. Clinic letters were searched from a database looking for specific key words to capture any patients who may have been missed in the logbook. Patients were excluded if they had pre-existing epiphora due to other causes or if marsupialisations were done for any reason other than related to malignant tumour excision. Failure was defined as any degree of epiphora following marsupialisation that was not secondary to any other cause, such as lid malposition.

Surgical method

Marsupialisation of the canalicular stump is well described by Older.[12] It involves opening the superior posterior part of the canaliculus for about 3–4 mm, then suturing the edges of the opened mucosal lining to the external surface, in order to create a large opening that will sit in the lacrimal lake.[12] This technique was modified as follows: the entire remnant canaliculus was opened up as far as possible to maximise all that was left, and no stents were used.

Literature review

Few studies examined only those patients with canalicular/lacrimal system sacrifice for tumour margin clearance. To contextualise the results of this series, the literature was reviewed to identify the rate of epiphora in those patients who sustained lacrimal system injury during tumour resection.

We searched MEDLINE as follows: "((((eyelid reconstruction) OR ("Eyelid Neoplasms/surgery"[MAJR])) OR ("Lacrimal Apparatus/surgery"[MeSH])) OR ("Skin Neoplasms/surgery"[MAJR])) AND (medial eyelid tumour)" and "((((eyelid tumour) AND lacrimal apparatus) AND eyelid reconstruction)) AND epiphora". Abstracts were reviewed to find the relevant papers, which were then considered in detail. Reference lists of those papers and of relevant reviews were checked to identify any other relevant papers that had not already been identified. Studies where the lacrimal system was injured or sacrificed as part of tumour excision, and with rates of postoperative epiphora for those patients were included. Studies where it was unclear which parts of the lacrimal system were sacrificed were excluded. Studies where patients with lacrimal system injury/sacrifice for tumour removal were part of a larger cohort were only included if it was possible to determine how many of these patients experienced post-reconstruction epiphora; that is, if there was an overall epiphora rate given for the whole cohort without specifying how many occurred in those with lacrimal system injury/sacrifice, then these studies were also excluded. Non-English articles were excluded.

Table 1 Tumour and surgical characteristics

Characteristic	N	%
Laterality		
Left	12	54.5
Right	10	45.4
Histology		
BCC	19	86.4
BCC background trichoepithelioma	1	4.5
BCC background tricholemmoma	1	4.5
Tarsal conjunctival SCC	1	4.5
Primary tumour excision		
Mohs surgery	2	9.1
Full thickness excision and paraffin section	20	90.9
Reconstruction		
Direct closure±lateral canthotomy/ cantholysis	13	69.1
Flap(s)*	5	22.7
Full-thickness skin graft	3	13.6
Hughes flap and full-thickness skin graft	1	4.5

*Flaps included tarsoconjunctival graft with skin and muscle advancement flap, periosteal flap, and skin and muscle transposition from the upper lid
BCC, basal cell carcinoma; SCC, squamous cell carcinoma.

RESULTS

There were 22 eyes (22 patients). The mean age of the patients was 77.6 years (95% CI 72.4 years to 82.8 years, SD 11.7 years). The rate of postoperative epiphora was 9.1% (2/22). The median follow-up was 16.6 months (range 3.7–127.9 months). The tumour and surgical characteristics are summarised in table 1.

All tumour resections included the lower canaliculus, and the lower canalicular remnant was marsupialised at the time of lid reconstruction. One patient additionally had a tumour in the lateral third of the upper lid, which did not involve the canalicular or lacrimal system. Thus, no superior canaliculi were marsupialised, and no superior canaliculi were resected.

Two patients experienced epiphora after lid reconstruction with marsupialisation of the remnant distal inferior canaliculus. One patient was a female in her 40s who had a right lower lid basal cell carcinoma on the background of a trichilemmoma which was excised and reconstructed with direct closure and marsupialisation of the inferior canaliculus. She initially reported minimal postoperative epiphora, which became 'troublesome' by 5.6 months postoperatively. At this point, the lower marsupialised system was not clearly visualised, and its functionality was questioned. To further assist this patient's symptoms, the upper system was assessed and a stenosed punctum was found. Sac washout was attempted, but cannulation of the punctum was not possible due to the severity of the stenosis. She underwent a right superior punctal dilation and three-snip punctoplasty procedure, which restored patency of the upper punctal system. A sac washout was done at the time of surgery through the upper punctum, and this demonstrated patency of the remaining lacrimal system. Review was planned following this, but she was lost to follow-up. Thus, it is unclear if this procedure alleviated her symptoms. She was then re-referred 8 months later to assess possible tumour recurrence in a new nodule under the scar from her previous surgery. Punch biopsy proved no malignancy. She continued to have troublesome epiphora. She subsequently received 5 units of botulinum toxin (BOTOX; Allergan plc, Dublin, Ireland) to the right lacrimal gland with good result and continued with this dose somewhat sporadically for five further treatments over the following 3 years. She wished for a more permanent solution, so concurrently, she underwent a conjunctivodacryocystorhinostomy with Lester Jones tube insertion and septoplasty for a deviated nasal septum as a joint procedure with otolaryngology. This was done 4.2 years following the original lid reconstruction and marsupialisation of the inferior canaliculus. She experienced a minor postoperative infection 1 week following her surgery, which resolved easily with oral co-amoxiclav and topical chloramphenicol ointment. At 1-month follow-up, the Lester Jones tube could no longer be seen in the eye nor visualised by nasoendoscopy in the clinic setting. She underwent surgical exploration with otolaryngology, and this showed the tube had migrated into her nose. Around this time, she received another 5 units of BOTOX to the right lacrimal gland. She underwent surgical repositioning of the tube, following a delay of 1 year and 10 months due to the COVID-19 pandemic. In the interim, she declined a top-up dose of BOTOX. At the time of writing this report, 5 months after the Lester Jones tube was repositioned, she has no symptoms of epiphora. She remains under review.

The other patient had epiphora noted to be 'not overly troublesome' at 6 months following lid reconstruction with direct closure. She was discharged from routine review at this point, 5 years and 7 months prior to writing this report. She has not re-presented.

Results of the literature review

The literature search yielded 284 results. The studies meeting the inclusion criteria are shown in table 2.

To facilitate direct comparison with this study, only those patients where the lacrimal system was injured/sacrificed for eyelid tumour removal were included in the table. Postoperative epiphora rates were calculated based on this. From these studies, the median epiphora rate was 12.5% (range 0%–100%).[10–12 14–29] The 100% was from a case report with a single patient.

DISCUSSION

The literature is heterogenous regarding the definitions of epiphora and of success. Some studies use the Munk grading scale to quantify the degree of epiphora,[10 16] which includes minor degrees of epiphora.[30] Others excluded

Table 2 Summary of the literature review

Study	Year published	Study type	N	Method of canalicular/lacrimal system reconstruction	Follow-up (months)	Epiphora N (%)	Comments
Laissez-faire/no reconstruction to lacrimal system							
Smit and Mourits[10]	1999	Retrospective case series	7*	Upper canaliculi intact	Not specified	3 (42.9)	▶ Only in cold and wind. ▶ Grade 1 and 2 Munk.
Meadows and Manners[17]	2003	Retrospective case series	1*	Glabellar or modified glabellar flap Upper and lower canaliculi excised	23	0 (0)	
Madge *et al*[18]	2010	Retrospective case series	20	Direct closure (1/20), complex reconstruction (19/20)	Median 25 Range 2–90	15 (75)	▶ Subsequent lacrimal bypass tube insertion, 4/20. ▶ Unknown severity.
Onaran *et al*[19]	2011	Case report	1	Paramedian forehead flap	8	1 (100)	Mild
Kesiktas *et al*[14]	2015	Retrospective case series	11	Glabellar rotation+nasolabial V-Y advancement flap	6	9 (81.8)	▶ Unknown severity. ▶ Lagophthalmos, 4/9.
Marsupialisation							
Older[12]	1979	Retrospective case series	3	Marsupialisation+silicone stent	24–36	0 (0)	Unknown severity/ quantification
Holds and Anderson[20]	1993	Retrospective case series	29	Marsupialisation following medial cantholysis for central/lateral lid tumours	≥6	1 (3.4)	Intermittent epiphora
Stent							
McCord[21]	1980	Retrospective case series	22*	Remnant canalicular stump externalised ('-ostomy' manner)+silicone stent	Not specified	0 (0)	Four overall (N=31) required canaliculostomy repositioning
Harrington[22]	1982	Retrospective case series	19*	Silicone stent or Veirs' rod, most bicanalicular	Range 2–90	5 (26.3)	▶ Minimal or slight epiphora, 4/19. ▶ Only in cold and wind, 1/19.
Lindgren *et al*[23]	2000	Prospective cohort	15*	Silicone stent (6/15)	Median 48 Range 3–120	9 (60)	▶ Surgery to improve lacrimal drainage failed, 4/9. ▶ Minor, no further surgery, 5/9.
Perry and Allen[15]	2016	Retrospective case series	8*	Crawford tube	Mean 5.6 Range 4–16	1 (12.5)	Continued epiphora despite patent lacrimal system
van Burink *et al*[16]	2018	Retrospective case series	10*	Mini-Monoka stent, sutured	3	1 (10)	Grade 1 Munk
Park and Kim[24]	2020	Case report	1	Remnant canaliculus transposed+Mini-Monoka (not specified if sutured)	12	0 (0)	Good tear drainage on dacyroscintigraphy at 6 months
Various							

Continued

Table 2 Continued

Study	Year published	Study type	N	Method of canalicular/lacrimal system reconstruction	Follow-up (months)	Epiphora N (%)	Comments
Lowry *et al*[25]	1997	Retrospective case series	3*	► Silicone stent (2/3). ► Marsupialisation (1/3).	► 60, 84. ► 7.	0 (0)	No evidence of nasolacrimal obstruction by final examination
Motomura *et al*[26]	2006	Retrospective case series	3	► Laissez-faire (2/3). ► Jones tube (1/3).	Mean 20 Range 12–25	2 (66.7)	Laissez-faire reconstruction, only with crying/in wind
Morton[11]	2016	Retrospective case series	18*	Marsupialisation if possible, or laissez faire (not specified how many in each)	Not specified	0 (0)	None with 'excess watering'
Yazici *et al*[27]	2021	Retrospective case series	14*	► Laissez-faire (12/14). ► Silicone stent (2/14).	Median 19 Range 1–91	3 (21.4%)	► 'Persistent epiphora'. ► All three bicanalicular involving.
Other							
Zapala *et al*[28]	1992	Retrospective case series	9*	Conjunctivorhinostomy, conjunctivodacyrocystorhinostomy, conjunctivosinusotomy	Not specified	6 (66.7)	► Partial obstruction, periodic epiphora worse outdoors (6/9). ► Failure (3/9).
Parker *et al*[29]	2014	Retrospective case series	3	Paramedian forehead flap with conjunctivorhinostomy using an AlloDerm as a conduit material	12, 18 and 13	0 (0)	Initially minimal epiphora in 2/3, but spontaneously resolved by last follow-up

*Only those within the study with medial lid tumours requiring sacrifice of any part of the lacrimal system for tumour margin clearance are included in this table.

epiphora if it was 'not to excess'[11] or conversely included it but quantified it by stating it was 'not interfering with daily life'.[13] Others did not quantify the degree of epiphora, making it difficult to compare to studies which did.[12 14 15]

It is well documented that both the upper and lower canaliculi are functionally important for tear drainage, and that one functioning canaliculus may be sufficient to prevent symptomatic epiphora, particularly in the absence of reflex tearing.[10 31–35] In this series, all cases involved the lower canaliculus, with all upper canaliculi remaining intact. The lack of epiphora symptoms could be attributed to the intact upper canaliculus. Hence, a comparative series, where the remnant canaliculi were not marsupialised, would have been very valuable. This was not possible, as it is not the practice in this unit. However, the literature contains studies where a laissez-faire approach is used, and no canalicular or lacrimal system reconstruction is attempted.

The rate of epiphora in these cases using a laissez-faire approach ranged from 42.9% to 81.8%,[10 14 18 26 27] if the reports where there was only one patient in the cohorts are excluded[17 19] (table 2). The series by Morton[11] and by Yazici *et al*[27] also used the laissez-faire approach to lacrimal reconstruction in addition to marsupialisation[11] and silicone intubation[27] but did not specify which method of lacrimal reconstruction was used in their patients with epiphora.

Of these series using the laissez-faire approach, Smit and Mourits,[10] (n=7) was comparable to this series, as none of the upper canaliculi were injured, and they reported on all epiphora, not only that which was problematic for the patient.[10] Their rate of epiphora of 42.9%[10] is higher than the rate found in this study of 9.1%. This suggests that marsupialisation of the remnant inferior canaliculus substantially aids in tear drainage, in addition to that which is drained by the intact upper canaliculus. Kesiktas *et al*,[14] Motomura *et al*[26] and Madge *et al*[18] also reported high rates of epiphora. The patients in Madge *et al*[18] underwent en bloc resection of the tumour, including the lacrimal sac, and Kesiktas *et al*[14] and Motomura *et al*[26] did not specify which parts of the lacrimal apparatus were removed during tumour excision but, based on the representative photos included, it is likely that both canaliculi were affected, if not much or all of the lacrimal apparatus. These may account for their much higher epiphora rates. Even in a more recent series (n=14), where the laissez faire approach was used in the majority of cases, with stenting in the others, the rate of epiphora was 21.4%, which was still higher than that in this series, and they only noted 'persistent epiphora' instead of all degrees of epiphora.[27]

For the most part, the epiphora reported in this context is mild (table 2), with a minority of patients requiring further surgery or intervention. In this study, one patient required extensive intervention for her epiphora symptoms. Perhaps this was due to her young age (46.1 years vs a mean of 77.6 years). It is well known that with increasing age, reflex tear secretion decreases.[36] Malignant eyelid tumours are more common in older patients, with the mean age of incidence of basal cell carcinomas and squamous cell carcinomas peaking in the seventh decade, and in the seventh to eight decades for sebaceous gland carcinomas.[37]

Due to the retrospective nature of the study, it was not possible to apply a uniform grading system to quantify the degree of epiphora nor were the results of syringing of the lacrimal system postoperatively universally available. The authors acknowledge these are limitations. However, it has previously been found that the presence or absence of epiphora is a more important and sensitive marker of the success of treatment for canalicular laceration, rather than an anatomically intact canalicular system.[10 38] Therefore, while anatomical success is academically interesting, using symptomatic epiphora as an endpoint is more useful pragmatically, particularly when considering the impact on the patient and on the health service.

No patients were lost to follow-up. In uncomplicated cases, those undergoing excision and reconstruction of lid tumours are routinely discharged from the unit 3–4 months afterwards. The authors acknowledge that 3–4 months is a relatively short amount of time. However, long-term patient outcomes were sought as much as feasibly possible within the limits of a retrospective study covering a large time frame and including the fact that half of the patients in the cohort were deceased by the time of data collection. It is unlikely that epiphora occurring after discharge from the unit was missed. The unit is one of four British ocular oncology centres. Hence, any subsequent ocular symptoms following treatment for ocular malignancy done in this unit, including epiphora, would be re-referred to the unit, irrespective of time elapsed. Furthermore, unrelated lid symptoms are typically re-referred to the unit, particularly as other care providers (primary care or local ophthalmology departments) prefer to have reassurance that the new symptom does not represent recurrence of the previously treated ocular malignancy.

Therefore, case notes were reviewed for reattendances or re-referrals following discharge. The nature of these was noted specifically for symptoms of epiphora, even if the patient had been referred or attended for something unrelated (eg, glaucoma screening). This was at a minimum review time of 2 years and 2 months but extending as long as 10.5 years. In the latter case, that patient is still under review due to the original malignancy being a tarsal conjunctival squamous cell carcinoma. A lack of reattendance or re-referral was taken to confirm a lack of symptomatic epiphora. The authors acknowledge that a lack of reattendance or re-referral does not equate to the absence of epiphora. However, the severity of epiphora is relevant when considering the impact on the patient. Mild or minor symptoms are tolerated by patients, without seeking further intervention.

The advantages of using marsupialisation in this setting are that it is a relatively straightforward procedure to

add onto the reconstructive surgery without excessively extending surgical time or recovery for the patient. The patient does not need to undergo an additional procedure for this to be done and, in doing so, may help the patient avoid secondary nasolacrimal rehabilitative procedures, which may be extensive and involve longer patient recovery. There is also no additional cost, which would incur when using a stent. The current cost to the department for one unit each of Mini Monoka (FCI Ophthalmics, Pembroke, Masssachusetts, USA), bicanalicular Crawford tubes (FCI Ophthalmics) and monocanalicular Crawford tubes (FCI Ophthalmics) is £81.67, £47.50 and £98.33, respectively. Furthermore, there is no risk of iatrogenic damage to the intact upper canaliculus or the remnant healthy canaliculus, as may occur when using stents. Morton[11] surmised his low rate of epiphora was due to leaving the remnant healthy canaliculus alone and therefore avoiding unnecessary trauma. There is also no upkeep required by the patient, as would be required if a Lester Jones tube was placed.[39] Moreover, this option remains as a fall back if marsupialisation fails. Therefore, marsupialisation of the remnant canaliculus during delayed reconstruction is a straightforward and effective surgical option that may help prevent postoperative epiphora when the proximal lacrimal system is sacrificed for tumour margin clearance.

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