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

Stephanie J Chiu, Zanna I Currie, Jennifer HY Tan

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.

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
to trauma, tumour resection, or punctal or canalicu-
lar stenosis or obstruction. For punctal stenosis/obstruc-
tion, treatment methods include perforated punctal
plugs, punctal snip procedures, punctal marsupialisa-
tion or recreation of a punctal opening. These may be
combined with recanalising procedures for the canali-
culus. For restoring canalicular patency, these methods
include balloon canaliculoplasty, canaliculare trephine,
and monocanalicular or bicanalicular stenting. Bypassing
the canaliculus and lacrimal sac altogether with a conjunc-
tivodacryocystorhinostomy and Lester Jones tube place-
ment remains the gold standard when there is extensive
canalicular obstruction, congenital agenesis, failed previ-
ous canalicular surgery or functional epiphora. 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 reconstruc-
tion and only reconstructing the lid defect, or leaving the
lid defect to heal by secondary intention.

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 retro-
spective 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. Marsupialisation of the remnant canalicu-
lus is a relatively straightforward procedure. It can be
easily incorporated into the reconstruction of the eyelid
following tumour resection. If successful, it would obvi-
ate 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 canalicu-
lus 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 Clin-
cal 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 conфи-
dential 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 canalicual 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 canalicual stump is well
described by Older. 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. 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 cana-
licular/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 recon-
struction) OR (“Eyelid Neoplasms/surgery”[MAJR])
OR (“Lacrimal Apparatus/surgery”[MeSH]) OR (“Skin
Neoplasms/surgery”[MAJR]) AND (medial eyelid
tumour)” and “(((eyelid tumour) AND lacrimal appa-
ratus) 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 iden-
tified. 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.
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 canalicus, 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 canalicus. She initially reported minimal postoperative epiphora. She subsequently received 5 units of botulinum toxin (BOTOX; Allergan plc, Dublin, Ireland) to the right lacrimal gland. 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 canalicus.

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

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, which includes minor degrees of epiphora. Others excluded...
| 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              | 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           | 2003           | Retrospective case series | 1* | Glabellar or modified glabellar flap                 | Upper and lower canaliculi excised | 23             | 0 (0)                                                                    |
| Madge et al                   | 2010           | Retrospective case series | 20 | Direct closure (1/20), complex reconstruction (19/20) | Median 25          | 15 (75)        | Subsequent lacrimal bypass tube insertion, 4/20. Unknown severity.       |
| Onaran et al                  | 2011           | Case report               | 1  | Paramedian forehead flap                             | 8                  | 1 (100)        | Mild                                                                      |
| Kesiktas et al                | 2015           | Retrospective case series | 11 | Glabellar rotation+nasolabial V-Y advancement flap   | 6                  | 9 (81.8)       | Unknown severity. Lagopthalmos, 4/9.                                      |
| Marsupialisation              |                |                           |    |                                                     |                    |                |                                                                            |
| Older                         | 1979           | Retrospective case series | 3  | Marsupilation+silicone stent                         | 24–36              | 0 (0)          | Unknown severity/quantification                                          |
| Holds and Anderson            | 1993           | Retrospective case series | 29 | Marsupialisation following medial cantholysis for central/lateral lid tumours | ≥6                 | 1 (3.4)        | Intermittent epiphora                                                    |
| Stent                         |                |                           |    |                                                     |                    |                |                                                                            |
| McCord                        | 1980           | Retrospective case series | 22*| Remnant canalicul stump externalised (‘-ostomy’ manner)+silicone stent | Not specified      | 0 (0)          | Four overall (N=31) required canaliculostomy repositioning               |
| Harrington                    | 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                | 2000           | Prospective cohort        | 15*| Silicone stent (6/15)                                | Median 48          | 9 (60)         | Surgery to improve lacrimal drainage failed, 4/9. Minor, no further surgery, 5/9. |
| Perry and Allen               | 2016           | Retrospective case series | 8* | Crawford tube                                        | Mean 5.6           | 1 (12.5)       | Continued epiphora despite patent lacrimal system                        |
| van Burink et al              | 2018           | Retrospective case series | 10*| Mini-Monoka stent, sutured                           | 3                  | 1 (10)         | Grade 1 Munk                                                           |
| Park and Kim                  | 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                       |                |                           |    |                                                     |                    |                |                                                                            |
| Study                  | Year published | Study type         | N   | Method of canalicular/lacrimal system reconstruction                      | Follow-up (months) | Epiphora N (%) | Comments                                                                 |
|------------------------|----------------|--------------------|-----|--------------------------------------------------------------------------------|--------------------|----------------|---------------------------------------------------------------------------|
| Lowry et al²⁵          | 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 al²⁶       | 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                      |
| Morton¹¹               | 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 al²²         | 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 al²⁸         | 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 al²⁹         | 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 exceed’ or conversely included it but quantified it by stating it was ‘not interfering with daily life’. Others did not quantify the degree of epiphora, making it difficult to compare to studies which did.

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. 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 comparative series, where the remnant canaliculi were involved the lower canaliculus, with all upper canaliculi 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 canicular 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%. If the reports where there was only one patient in the cohorts are excluded, if the reports where there was only one patient in the cohorts are excluded, the rate of epiphora of 42.9% 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., Motomura et al.26 and Madge et al.28 also reported high rates of epiphora. The patients in Madge et al.28 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.

For the most part, the epiphora reported in this context is mild, 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. 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 eighth decades for sebaceous gland carcinomas.

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 canicular laceration, rather than an anatomically intact canicular system. 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, Massachusetts, 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. Morton11 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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ORCID iD Stephanie J Chiu http://orcid.org/0000-0003-0932-4864

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