

## External dacryocystorhinostomy for isolated lacrimal sac rhinosporidiosis – A suitable alternative to dacryocystectomy

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**Purpose:** To describe the outcome of external dacryocystorhinostomy (DCR) as a suitable alternative to dacryocystectomy (DCT) in cases of isolated lacrimal sac rhinosporidiosis. **Methods:** This was a retrospective, interventional case series. Institutional review board approval was obtained for the study. The chart review of 13 patients who underwent external DCR surgery for isolated lacrimal sac rhinosporidiosis between July 2012 and May 2018 was performed. Demographic details, clinical presentation, preoperative and intraoperative findings, surgical technique used, postoperative management, surgical outcome and duration of follow up were reviewed for each patient. **Results:** Thirteen patients were included in the study. Nine (69.2%) were male and four (30.8%) were female. The mean age was 22.1 years (range: 8–46 years). Involvement was unilateral in all cases. The commonest presenting complaint was epiphora with discharge seen in 10 cases (76.9%). An intrasac granuloma was grossly identified intraoperatively in 12 out of the 13 patients. Mean follow up was 26.9 months (range: 1.5–68 months). Till the last follow up, all cases were symptom free and did not have any evidence of recurrence of infection. **Conclusion:** From this series of cases reported by the authors, external DCR with appropriate precautions to prevent recurrence appears to be a suitable alternative to conventional DCT with excellent long-term outcomes without disease recurrence.

**Key words:** External dacryocystorhinostomy, lacrimal sac, rhinosporidiosis

Rhinosporidiosis, a chronic granulomatous infection caused by *Rhinosporidium seeberi*, most commonly affects mucosal surfaces and has been reported in the literature to involve diverse sites in the body including the nasal cavity, nasopharynx, oropharynx, the upper respiratory tract, the ocular surface (most commonly the conjunctiva but rarely causing scleral thinning), the lacrimal drainage system (primarily the lacrimal sac), the genitalia, the rectum, the skin and very rarely even the bone. The commonest site reported in the ocular and adnexal region is the conjunctival surface (involved in >90% of reported cases)<sup>[1,2]</sup> followed by the lacrimal sac (involved in about 5–24% of cases as reported in different series).<sup>[1–3]</sup> The causative agent of the disease, *Rhinosporidium seeberi*, is currently believed to be a eukaryotic parasite and has eluded growth in cultures.<sup>[4,5]</sup> The objective of this study was to report the outcomes of external dacryocystorhinostomy (DCR) as the treatment modality of choice as an alternative to the conventionally recommended and more commonly performed dacryocystectomy (DCT) for cases with isolated lacrimal sac rhinosporidiosis. To the best of the authors knowledge, only one other report is available in the ophthalmic literature that describes this surgical approach for lacrimal sac rhinosporidiosis.<sup>[6]</sup>

### Methods

This study involved a retrospective patient chart review. Institutional review board approval was obtained and the

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tenets of the Declaration of Helsinki were followed during the conduct of the study. Patients who underwent external DCR surgery for nasolacrimal duct obstruction between July 2012 and May 2018 were reviewed and those diagnosed to have rhinosporidiosis confirmed on histopathological examination of sac tissue were included in the study. A total of 13 consecutive patients were found suitable for inclusion in the study. Demographic details, clinical presentation, preoperative and intraoperative findings, surgical technique used, postoperative management, surgical outcome and duration of follow up were reviewed for each patient. The surgical technique used for external DCR was similar to the one previously described in the literature.<sup>[6]</sup> Apart from the routine steps of an external DCR surgery, the specific measures taken for prevention of recurrence of infection were as follows – the lacrimal sac fossa was treated with an application of 5% povidone iodine solution for 5 min intraoperatively after incising the lacrimal sac, excising the intrasac granuloma and fashioning the sac flaps. The posterior sac flap was then excised and the anterior sac flap was trimmed before anastomosis with the nasal mucosal flap. No cauterisation was used for the surrounding tissue in an attempt to minimise the risk of scarring in the region of the internal common canalicular opening; an intranasal povidone iodine-soaked pack was left *in situ* at the conclusion of surgery

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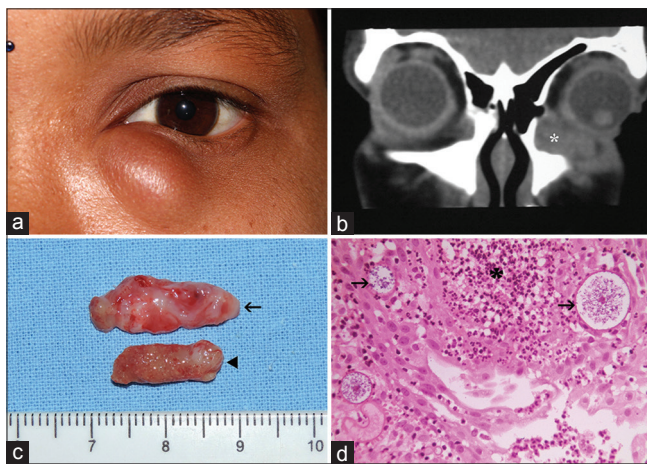
and was removed 12 h postoperatively. Decision to perform an intraoperative bicanalicular silicone intubation was left to the surgeon's discretion. Postoperative follow up was at 1 week, 6 weeks, 3 months and 6 months. Thereafter, follow up was yearly. At each visit, symptoms related to lacrimal drainage function were documented and lacrimal passage irrigation was performed. The incision wound site was checked and the nasal cavity was evaluated endoscopically in detail (under topical anaesthesia application) for possible disease recurrence. For the patients who received intubation, tube removal was performed 6 weeks postoperatively. A successful outcome for the DCR was defined by the absence of lacrimal symptoms and the presence of a freely patent ostium on lacrimal irrigation at each follow-up visit.

## Results

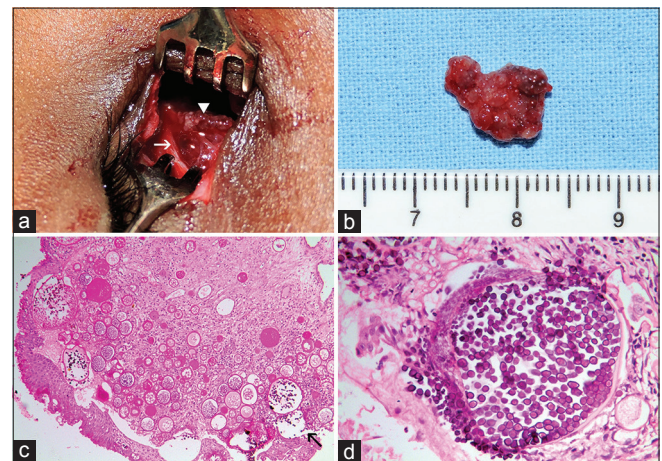
Of the 13 patients included in the study, 9 (69.2%) were male and 4 (30.8%) were female. The mean age was 22.1 years (range 8–46 years) with six patients (46%) being <15 years of age. The right side was affected in six cases and the left side in the other seven cases. Involvement was unilateral in all cases. The commonest presenting complaint was epiphora with discharge seen in 10 cases (76.9%) followed by the presence of a swelling in the region of the lacrimal sac noted by the patient in 7 cases (53.8%). In only two of these patients, the sac swelling had a doughy feel on palpation. A concurrent lower eyelid swelling was reported by two patients and in one of these patients the swelling had a doughy feel on palpation. None of the cases reported to have noticed the presence of blood associated with epiphora/discharge at any point during the time lag from symptom onset to presentation. This lag time ranged from as early as 2 weeks to as late as 2 years.

A definite history of either regular or occasional bathing in stagnant pond water was elicited in 7 (53.8%) of the

13 cases. The best-corrected visual activity was 20/20, N6 in the affected eye for all patients except for one who had a best-corrected visual acuity of counting fingers close to the face in the affected eye due to the presence of a dense cataract. Regurgitation of mucoid, mucopurulent or frank purulent material on pressure applied over the lacrimal sac region was noted in 10 patients. None of the cases had a haemorrhagic regurgitation. One of these patients had a patent lacrimal system on syringing and had an atonic sac that decompressed through the nasolacrimal duct into the nasal cavity by pressure application over the sac with punctal occlusion. Of the remaining three patients, one had an encysted mucocele, one had a non-patent lacrimal passage on syringing and the third had a patent lacrimal system on syringing. One of the cases had a history of surgical excision of a left-sided nasal polyp diagnosed to be rhinosporidiosis but had not had any recurrence on follow up. The rest of the cases had no evidence of nasal involvement with rhinosporidiosis on preoperative evaluation. An intrasac reddish, granulomatous, friable soft tissue mass was grossly identified intraoperatively [Fig. 1] in all but one of the cases. In this one case, the sac wall appeared to be grossly thickened. One of the cases also had a sac diverticulum that extended inferolaterally from the body of the sac [Fig. 2]. An external DCR with bicanalicular silicone intubation was performed in nine cases and an external DCR without intubation was performed in the remaining four cases. There was profuse intraoperative haemorrhage resulting from the intrasac granuloma in four of the cases. In one of these, a non-endoscopic endonasal DCR was started initially but was abandoned midway due to obscured visibility caused by the profuse intraoperative haemorrhage and was eventually converted into an external DCR procedure. Postoperative recovery was uneventful in all cases. Histopathological confirmation of lacrimal sac rhinosporidiosis was found in all the cases. Postoperative oral dapsone was prescribed in five



**Figure 1:** This patient presented with a medial lower lid swelling (a) and watering from the left eye. Computed tomography showed a soft tissue lesion corresponding to the swelling (b, asterisk). At surgery, a sac diverticulum (c, black arrow) and an intrasac granuloma were found. The granuloma had multiple sago-grain like sporangia visible on its surface (c, black arrowhead). On light microscopy (H and E, 40 $\times$ ), squamous metaplastic epithelium of sac with intraepithelial sporangia filled with endospores (d, arrows) was noted. The stroma was oedematous with a mixed inflammatory response and a neutrophilic abscess (d, asterisk)



**Figure 2:** An intrasac granuloma was noted in this patient intraoperatively (a, arrow shows border of incised sac wall, arrowhead shows the granuloma) and was found to have multiple sago-grain like sporangia visible on the surface (b). On light microscopy (c, PAS, 10 $\times$ ), stratified columnar epithelium of the lacrimal sac with focal thinning and denudation was noted (c). The stroma was diffusely oedematous and showed numerous sporangia of varying sizes and stages of maturation with some filled with endospores (c). Rupture of sporangia with release of the endospores was also evident (c, arrow, d, H and E, 40 $\times$ )

cases. Mean follow up was 26.9 months (range: 1.5–68 months) with nine of the cases (69.2%) having a follow up of 6 months or more. Till the last follow up, all the cases were symptom free, had patency on lacrimal passage irrigation and did not have any evidence of recurrence of rhinosporidiosis. The results have been summarised in Table 1.

## Discussion

Rhinosporidiosis is caused by *Rhinosporidium seeberi*, first described by an Argentinian physician Guillermo Seeber in 1900,<sup>[7]</sup> and is known to be endemic on the Indian subcontinent, Sri Lanka, Bangladesh and Nepal.<sup>[6,8]</sup> However, it has been reported from diverse geographical locations worldwide.<sup>[8]</sup> Infection is commoner in the first four decades and is 2.5 times commoner in men.<sup>[2]</sup> About 15% of cases involve the eye and the ocular adnexa most commonly affecting the conjunctiva followed by the lacrimal sac.<sup>[2,8]</sup> Bathing in stagnant pond water is believed to be a definite risk factor in contracting the disease.<sup>[8]</sup> Infection has also been postulated to be acquired by inhalation of contaminated field dust.<sup>[9]</sup> The organism is believed to thrive in a humid, warm, tropical climate and stagnant pond water is believed to provide an optimal environment for its survival.<sup>[10]</sup>

The choice of treatment for rhinosporidiosis at any involved site is complete local excision of the granulomatous lesion with adequate cauterisation. Spillage and seeding of the spores on adjacent normal tissues is to be prevented to avoid possible recurrences.<sup>[8]</sup> The lacrimal sac being a relatively isolated organ compared to the other involved sites in the body, lends itself to complete removal more easily than most other tissues. It is therefore not surprising that the most commonly recommended treatment modality for lacrimal sac rhinosporidiosis in the literature is complete excision of the infected lacrimal sac or DCR.<sup>[1–3,11,12]</sup> This modality, though apparently safe and curative, also produces constant and debilitating postoperative epiphora in the patient that is difficult to relieve. In contrast, a DCR performed after excision of the sac granuloma with appropriate precautions is a suitable alternative that appears to have very good long-term outcomes

as suggested by the authors' study. There are several clinical pointers that raise a suspicion of lacrimal sac rhinosporidiosis during clinical evaluation – a boggy lacrimal sac swelling with a 'bag of worms' feel,<sup>[3,11]</sup> presence of bloody tears,<sup>[13]</sup> history of bathing in stagnant pond water<sup>[1,10]</sup> and residence in or recent travel to an endemic area.<sup>[2,8,10]</sup> In the current study, four patients (30.8%) had a boggy lacrimal sac and none had bloody tears. Seven cases (53.8%) had a history of bathing in stagnant pond water and six (46.2%) were residents of an endemic geographic area. A male preponderance was noted (2.3:1) and almost half (46%) the affected cases were <15 years of age. These features matched well with the socio-demographic correlates of rhinosporidiosis described in the literature.<sup>[14]</sup> Nuruddin *et al.* published the only other paper in the ophthalmic literature that describes a study similar to the current one.<sup>[6]</sup> They reported a series of 18 patients with the highest occurrence of infection in the 25–34 year age group (<15 years for the current study), a male to female preponderance of 3.5:1 (2.3:1 for the current study), a history of bathing in stagnant water in 66.7% (53.8% in the current study), a doughy lacrimal sac swelling in 100% (30.8% in the current study) and bloody discharge from the nose in 61.1% (none in the current study). Only 22.2% of their patients complained of epiphora as a presenting symptom as compared to 76.9% in the current study. It has been reported that epiphora is not a common symptom in lacrimal sac rhinosporidiosis as the spread of infection tends to be pericanalicular and perisaccular,<sup>[15]</sup> but this was not seen to be true for the current study. In fact, an intrasac granuloma was identified at surgery in 12 patients (92.3%) in the current study.

The follow up reported by Nuruddin *et al.* in their case series was for a minimum of 12 months.<sup>[6]</sup> The maximum follow up was not mentioned. They reported a recurrence in two cases (11.1%) in the sac region within 2 months of surgical intervention. They believed that intraoperative application of a 5% povidone iodine gauze to the lacrimal fossa for 2 min helped in prevention of recurrence. The current study had a mean follow up of 26.9 months and had no recurrences in

**Table 1: Patient demographics, presentation, treatment and outcome in isolated lacrimal sac rhinosporidiosis**

Patient No.	Age	Sex	Clinical presentation/ laterality	History of bathing in ponds	Surgery performed	Postoperative oral dapsone prophylaxis (months received)	Follow up (months)	Outcome
1	12	F	SS, LLS, W, ROP/R	Y	DCR + TUBE	Y (3)	68	S/NR
2	24	M	SS, W, D, ROP/L	N	DCR	Y (3)	52	S/NR
3	11	M	W, D, ROP/L	Y	DCR + TUBE	Y (3)	25	S/NR
4	43	F	SS, ROP, ATS/R	Y	DCR + TUBE	Y (3)	3	S/NR
5	35	M	SS, EN-MUC/R	N	DCR	Y (3)	34	S/NR
6	30	M	SS, ROP/L	N	DCR (EN-EXT)	N	66	S/NR
7	14	M	W, D, ROP/L	N	DCR	N	54	S/NR
8	28	M	SS, W, D/L	N	DCR + TUBE	N	27	S/NR
9	8	M	SS, W, D, ROP/R	Y	DCR + TUBE	N	9	S/NR
10	9	M	W, D, ROP/R	Y	DCR + TUBE	N	7	S/NR
11	46	F	W, D, ROP/L	N	DCR + TUBE	N	4	S/NR
12	9	M	W, D, ROP/R	Y	DCR + TUBE	N	3	S/NR
13	18	F	LLS, W/L	Y	DCR + TUBE	N	3	S/NR

M=Male, F=Female, SS=Sac swelling, LLS=Lower lid swelling, W=Watering, D=Discharge, ROP=Regurgitation on pressure over the sac, R=Right, L=left, ATS=Atonic sac, EN-MUC=Encysted mucocele, DCR=Dacryocystorhinostomy, EN-EXT=Endonasal to external, S=Successful, NR=No recurrence, Y=Yes, N=No



any of the cases. The authors also used 5% povidone iodine solution but they completely filled the lacrimal sac fossa for 5 min and followed it up with a povidone iodine soaked nasal pack left in contact with the osteotomy site for 12 h postoperatively. This may have possibly contributed to more effective prevention of recurrence of infection. Povidone iodine is reported to cause metabolic inactivation of endospores with an exposure time of approximately 7 min.<sup>[16]</sup> It has also been recommended for use in nasal packs<sup>[16]</sup> as was done by the authors in the current study.

Dapsone has been reported as a useful agent for medical management of rhinosporidiosis and has been recommended by some authors for a period of 1 year.<sup>[17,18]</sup> It is believed to be effective in tackling local subepithelial and subcutaneous spread<sup>[17]</sup> and acts by causing maturation arrest of sporangia and accelerates their degeneration. These non-dividing sporangia are removed by an accentuated granulomatous response.<sup>[18]</sup> There are no specific guidelines in the literature about postoperative medical prophylaxis with dapsone for recurrence in patients undergoing surgery for isolated lacrimal sac rhinosporidiosis. Nuruddin *et al.* prescribed long-term dapsone to two cases in their series but only after the development of postoperative recurrence of infection and not as prophylaxis. They did not specify the exact duration that the drug was administered for.<sup>[6]</sup> In the current study, five (38.5%) cases received postoperative prophylaxis with oral dapsone for a period of 3 months. These were the first five cases of the series. Subsequently, three patients refused long-term prophylaxis with dapsone but did not have any disease recurrence on long-term follow up. Based on this observation, the authors did not advise routine postoperative dapsone for the remaining patients.

## Conclusion

From this reported series, external DCR appears to be a suitable alternative to the more conventionally performed DCT for cases of isolated lacrimal sac rhinosporidiosis. Clinical pointers as enumerated previously are key in raising suspicion of the presence of sac rhinosporidiosis during preoperative evaluation. DCR performed in such cases with appropriate precautionary measures can lead to excellent long-term outcomes with no recurrence of infection. With regard to postoperative dapsone prophylaxis, the patient numbers in this study are too small to be analysed meaningfully. However, based on the experience gained till date, the authors currently do not use routine dapsone prophylaxis for these cases in their own practice.

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## Conflicts of interest

There are no conflicts of interest.

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