• Clinical Research •

Behavior and outcomes of 141 acquired lacrimal sac mucoceles treated *via* endoscopic dacryocystorhinostomy

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Abstract

● **AIM:** To investigate the clinical profile of patients with acquired lacrimal sac mucocele (ALSM) and evaluate the efficacy of endoscopic dacryocystorhinostomy (En-DCR) for this condition.

● **METHODS:** En-DCRs were performed on 141 patients with ALSM patients from January 2016 to March 2022. The clinical baseline information and magnetic resonance imaging (MRI) images were recorded and summarized. To assess the effectiveness of En-DCR therapy, both anatomical and functional success rate was assessed during a 12mo follow-up.

● **RESULTS:** A total of 141 patients, with a mean age of 57.70±14.11y, were enrolled in this study. Majority of the patients were female (*n*=91; 64.54%) and all had unilateral disease. All patients had a previous history of epiphora and purulent secretion, and the duration from lacrimal duct obstruction to mucocele formation ranged from 6 to 120mo. MRI findings consistently revealed an enlarged sac diameter, fluid accumulation separated by a thin rim from adjacent tissues, which is indicative of lacrimal sac mucocele. En-DCR was performed with an anatomical success rate of 93.62% and a functional success rate of 81.56%.

● **CONCLUSION:** ALSM is more commonly seen in females and unilaterally. It is essentially a complication of lacrimal duct obstruction. MRI characteristics can be used for precise clinical diagnosis, while En-DCR emerges as an optimal therapy for this condition. Our results provide a comprehensive reference for the diagnosis and treatment of ALSM.

● **KEYWORDS:** acquired lacrimal sac mucocele; clinical profile; magnetic resonance imaging; endoscopic dacryocystorhinostomy

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INTRODUCTION

Lacrimal sac mucocele is characterized by the distension of the lacrimal sac, resulting in a bluish cystic swelling beneath the medial canthal area. Its main symptom is excessive tearing[1]. This condition commonly occurs in newborns due to congenital stenosis of the lacrimal system. The development of congenital lacrimal sac mucocele is primarily attributed to blockage at the valves of Rosenmuller and Hasner[2]. A similar mechanism may be involved in acquired lacrimal sac mucocele (ALSM), which arises from obstruction of the distal nasolacrimal duct caused by various etiologies and functional obstruction at the junction between common canaliculus and sac[1,3-4]. Magnetic resonance imaging (MRI) plays a crucial role in diagnosing lacrimal sac mucocele as it helps differentiate it from distinguish orbital- or naso-lacrimal sac tumors located in the medial canthal area and adjacent regions[5-6]. Although previous studies have reported on the effectiveness of dacryocystosinostomy for treating this condition, most were limited to case reports and small case series focusing on treatment outcome[1-2,4,6]. To address this gap, we conducted a comprehensive study involving 141 patients with ALSM, analyzing their clinical profile, natural history, MRI images, management using endoscopic

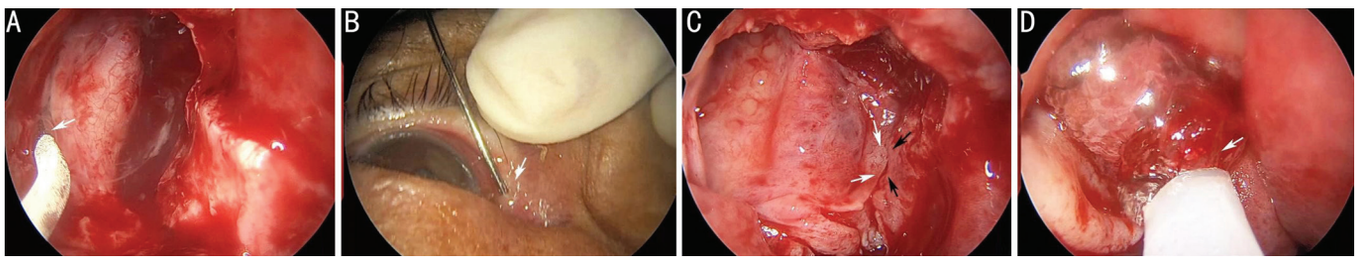

Figure 1 Intraoperative endoscopic views of the En-DCR in a patient with ALSM A: A 20-gauge microvitreoretinal ophthalmic knife was used to incise the inferior anterior portion of the lacrimal sac to release the purulent material therein (arrow); B: The medial wall of the sac was tented by inserting a Bowman probe through the superior punctum (arrow); C: The posterior lacrimal sac flap (white arrow) was made and then flattened to oppose the nasal mucosal flap (black arrow); D: Hyaluronic acid hydrogel was filled in the ostia (white arrow). ALSM: Acquired lacrimal sac mucocele; En-DCR: Endoscopic dacryocystorhinostomy.

dacryocystorhinostomy (En-DCR), and treatment outcomes. Our study provides a valuable clinical reference for diagnosing and managing ALSM.

PARTICIPANTS AND METHODS

Ethical Approval The present retrospective observational study was conducted at the Eye Hospital of Wenzhou Medical University in Zhejiang Province. This study was authorized by the Eye Hospital of Wenzhou Medical University and followed the principles outlined in the Helsinki Declaration (2024). It was approved by the Institutional Ethics Committee (2023-065-K-53; Medical Ethics Committee, Wenzhou Medical University, Wenzhou, Zhejiang Province, China). Written informed consent from all patients.

Patients presenting with a palpable mass at the medial canthal region of the lower eyelid, along with a soft ending upon lacrimal probe placement into the lacrimal canaliculi, were initial diagnosed with ALSM. All cases underwent comprehensive eye exam, endoscopic exam, and MRI assessment before En-DCR. MRI exam was performed using a with a 1.5 T machine (MAGNETOM ESSENZA) in the horizontal plane, and different radiologists measured the maximum dacryocyst diameters in two planes (horizontal and sagittal lengths in the horizontal plane and vertical length in the vertical plane). These data were recorded, including clinical profile, MRI images, En-DCR management details, outcomes, and complications. MRI was performed to further confirm the diagnosis and rule out other diseases, such as tumors and inflammatory pseudotumor. Patients with history of eyelid surgery and injury, eyelid abnormalities, facial paralysis, or less than 12mo follow-up were excluded. Fluorescein tests were conducted on 35 patients with ocular disorders. Briefly, fluorescein eye drops (5min a drop for 4 times) were added in the eyes 2 to 3h prior to surgery, and then the fluorescein determined present or not in the lacrimal sac at the time of incision.

All patients underwent En-DCR surgery with a large bony ostium and mucosal anastomosis of the posterior lacrimal sac flap and nasal mucosal flap (Figure 1)[7]. Silicone tubes intubation was not performed, except for cases with canaliculus stenosis.

Postoperative care regimen involved twice-daily administration of intranasal Rhinocort Aqua nasal spray (Astra Zeneca, Wilmington, USA) for 8wk in all subjects. Additionally, 0.5% levofloxacin and 0.02% fluorometholone (both from Santen Pharmaceutical Co, Ltd., Japan) were locally applied for 2wk. Follow-up was conducted at 1, 2wk, 1, 3, 6, and 12mo after En-DCR surgery. The symptoms, particularly epiphora and purulent discharge, were carefully recorded each follow-up. The Munk scale was used to subjectively assess the severity of epiphora[8]. Endoscopy with fluorescein exam was performed to assess the patency of the rhinostomy. Bicanalicular intubation was removed 3mo after surgery. Anatomical success was defined as the resolution of infection and a clear patent ostium on irrigation. Functional success was defined as the resolution of infection and absence or minimal presence of epiphora (Munk score was 0 or 1), along with unobstructed dye flow through the ostium during functional endoscopic dye testing[8-9].

Statistical Analysis Statistical analyses were performed using SPSS 26.0 software. The demographic data was compared using independent *t*-test or Chi-squared test. The success rates were compared using Pearson Chi-squared test or Fisher's exact test. Differences were considered significant when $P<0.05$.

RESULTS

A total of 159 patients (159 eyes) underwent En-DCR were included in this study, spanning over 5y ranging from Jan. 2016 to March 2022. However, 18 patients failed to follow-up. Finally, we included 141 patients with a mean age of 57.70±14.11y. Most of the patients were female (n=91; 64.54%) and all had unilateral disease. Among them, left lacrimal sac mucocele occurred in 65 cases (46.10%).

All the patients presented with chronic dacryorrhea prior to mucocele formation. Nasolacrimal duct obstruction (NLDO)

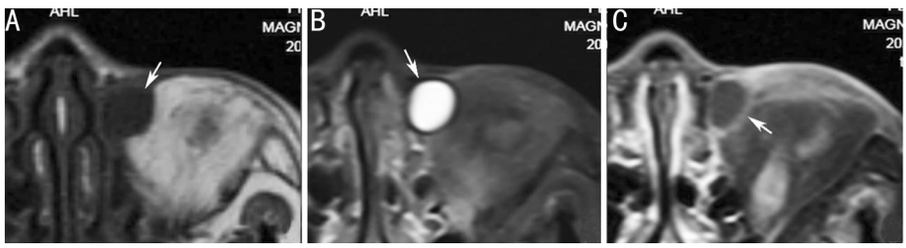

Figure 2 MRI imaging for ALSMs Axial MRI imaging with T1WI (A), T2WI (B), and contrast-enhanced T1WI (C) demonstrating a lacrimal sac mucocele (arrow). MRI: Magnetic resonance imaging; ALSM: Acquired lacrimal sac mucocele.

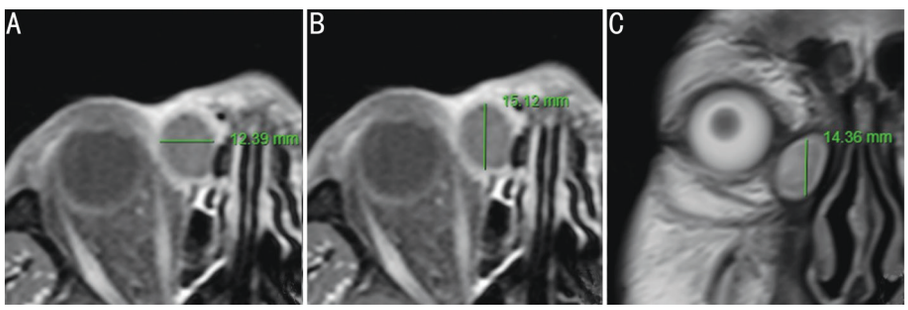

Figure 3 Maximum dacryocyst diameters in three planes being measured in MRI images A, B: Horizontal length and sagittal length were measured in sagittal planes; C: Vertical length was measured in vertical plane. MRI: Magnetic resonance imaging.

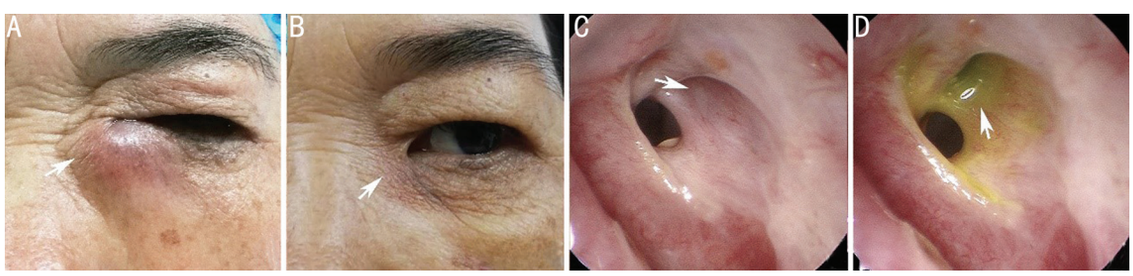

Figure 4 A case with ALSM before and after En-DCR A: A patient exhibited lacrimal abscess formation (arrow); B: The lacrimal abscess disappeared the first post-operation day (arrow); C: Endoscopic views revealing the left intranasal ostium remained patent and was covered by a mucosal layer exhibiting normal epithelial characteristics (arrow); D: Normal functional endoscopic dye test results (arrow). En-DCR: Endoscopic dacryocystosinostomy; ALSM: Acquired lacrimal sac mucocele.

was attributed to trauma in 21 cases and endoscopic operation in 7 cases, while the causes remained unknown in most cases. The average time from onset of lacrimal duct obstruction to mucocele formation was 41.94±21.69mo (ranging from 6mo to 120mo). Furthermore, the mean duration from mucocele formation to En-DCR surgery was 9.05±12.02mo (ranging from 1mo to 24mo).

In all cases, the main clinical signs and symptoms of ALSM were epiphora and swelling in the region of the lacrimal sac. For the clinical manifestation of swelling, 117 cases (82.98%) presented with mild symptoms in the morning that progressed to severe symptoms by evening. Out of the 35 cases who underwent a fluorescein test, 27 showed varying degrees of symptoms while 8 did not exhibit any such phenomena. During lacrimal sac incision, fluorescein was observed in the sac of all the 27 cases with these phenomena, whereas it was absent in the 8 cases without such symptoms.

The MRI analysis revealed a hypointense mass on T1-weighted images (Figure 2A) and a hyperintense mass on T2-weighted images (Figure 2B). Contrast enhancement in the walls of mucoceles was observed in all cases through enhanced MRI scans, while no internal contrast enhancement within the mass was detected (Figure 2C). Additionally, MRI demonstrated an increase in diameters for all cases (Figure 3), with horizontal length ranging from 6.83-12.39 mm (mean 9.05 mm), sagittal length ranging from 7.06-15.12 mm (mean 9.66 mm), and vertical length ranging from 8.64-16.01 mm (mean 11.43 mm). All patients received En-DCR therapy, resulting in a significant reduction in the size of the mass upon opening of the lacrimal sac. Complete resolution of swelling was achieved within 5d post-operation (Figure 4A, 4B). During the one-year follow-up, anatomical success was observed in 132 eyes (93.62%; Figure 4C, 4D). However, despite anatomical patency in these cases, epiphora persisted in 17 eyes with a Munk score ≥2, leading to an overall functional success rate of 81.56% (115/141). Anatomical failure occurred in nine cases: one case due to inferior canalicular obstruction and eight cases due to intranasal ostial closure–five attributed to scarring and

three attributed to granuloma formation. Six of the seven patients who underwent intubation procedures achieved both anatomical and functional success, yielding a success rate of 85.71%. In addition, bicanalicular intubations were required for seven cases (4.96%) due to canaliculus duct stenosis, and the bicanalicular intubation was removed three months after surgery.

The patients did not experience any major complications after surgery, such as significant nasal bleeding, infection spreading, or changes in vision, during the follow-up period.

DISCUSSION

A lacrimal sac mucocele is characterized by the swelling of a lacrimal sac, primarily resulting from distal nasolacrimal duct blockage[1-2]. The formation of lacrimal sac mucocele can be explained through two possible mechanisms. First, due to the NLDO, secretions may accumulate within the lacrimal sac distends, causing it to expend and exert pressure on the 2 canaliculi, leading to their folding upon themselves and subsequent proximal obstruction. Another postulated mechanism involves kinking of the common canaliculus because of a distended lacrimal sac and the potential dysfunction of Rosenmuller's valve–which serves as an entrance for the common canaliculus into the lacrimal sac–secondary to edema and inflammation[1,3,9].

ALSM is usually secondary to dacryocystitis, and dacryocystitis has various causes, but the common result is the complete obstruction of the nasolacrimal duct, thus resulting in stasis of tear flow, leading to secondary infections, which might progress to lacrimal sac mucocele[1,4]. In this study, all the enrolled patients presented with a symptom of epiphora and purulent secretion before the mucocele formation, which means that all patients had a history of NLDO and dacryocystitis. A variety of different mechanisms can contribute to obstruction of the distal passageways[10]. However, the specific causes of NLDO were unclear in most cases, and a part of them exhibited clear etiology, 21 of them were due to trauma, and 7 cases were due to endoscopic sinus surgery in the study.

Swelling in the lacrimal sac area and epiphora were the main symptoms associated with ALSM in all eyes[4-6]. Moreover, interesting phenomena were found in most patients. For instance, in 117 patients, lacrimal sac mucoceles were observed to be mild in the morning and severe in the evening. This clinical manifestation can be possibly explained by the pathogenesis of the disease, which have been described above. During the day, the fluid can ingress the dacryocyst by blinking, but limits egress thus leading to the dilation of the sac. It has been found that the mucous membrane of the dacryocyst absorbs the fluid in the dacryocyst during the night, thus causing the shrinkage of the sac[6]. However, in the other 24 patients, there was no such obvious manifestation. A postulated mechanism is that the pressure inside the lacrimal sac is high enough or the entrance of the common canaliculus into the lacrimal sac is 'severe obstruction' that can prevent the tears from entering to the lacrimal sac. In addition, the amount of fluid as well as the mucus secreted and absorbed by the lacrimal sac is kept in a balanced state. We did fluorescein tests with 35 patients in a non-randomized manner that confirmed our hypothesis. In 27 of these phenomena patients, fluorescein eye drops (5min a drop for 4 times) were added in the affected eyes 2 to 3h earlier on the day of surgery, and thereafter the fluorescein determined present in all lacrimal sac at the time of lacrimal sac incision. However, it was not found in 8 patients who lacked these phenomena.

En-DCR displayed an anatomical success rate of 93.62% and functional success rate of 81.56% in the study. The anatomical success rate was comparable to that of chronic dacryocystitis, but the functional success rate was significantly lower than in comparison to the previously reported studies[7,10-11]. Moreover, in prior reports related to the treatment of acquired dacryocyst, either functional epiphora was not evaluated, or only a few cases were reported[1,4]. Interestingly, in some large sample reports of En-DCRs of dacryocystitis, the possibility of functional epiphora has been reported in around 2% of cases[7,11]. However, functional epiphora was present in over 12% (93.62%-81.56%) of patients in the study. There could be two possible reasons for these observations: First, Munk score was used to evaluate the functional success and a score ≥2 was considered as functional failure in the study, which might effectively lower the functional success rate. Second, the higher functional failure could be potentially related to the mechanism of dacryocyst formation, and the dysfunction of the common canalicular by persistent tension in the dacryocyst may be the primary cause of the functional epiphora. We found that it can take some time from obstruction of nasolacrimal duct to lacrimal sac mucocele formation, and the mean duration of the symptom of lacrimal duct obstruction to the onset of cyst formation was 41.94±21.69mo (ranging from 6mo to 120mo). Therefore, timely treatment of NLDO or chronic dacryocystitis can effectively prevent the onset of ALSM and might improve the functional success rate. In addition, the mean duration of the cyst formation to the En-DCR surgery was 9.05±12.02mo (ranging from 1mo to 24mo). However, whether timely operation after the cyst formation can significantly improve the functional success rate is also a question worthy of further study.

Imaging examination is necessary to confirm the diagnosis because many tumors in or around the lacrimal sac can also lead to the local swelling. MRI examinations can reveal the occurrence of orbital tumors in the medial canthal region,

such as hemangioma, neurofibroma, lymphangioma and rhabdomyosarcoma, as solid soft-tissue masses with variable degrees of enhancement[1,5-6]. In this study, MRI was able to depict the margins of a mucoceles, its contents, mucosal thickening, retained secretions, retention cysts, and set apart the tumors from inflammation, which can effectively aid to differentiate between the various pathological soft-tissue lesions. MRI imaging revealed a potential hypointense mass on T1-weighted images and a hyperintense mass on T2-weighted images, whereas enhanced MRI examinations revealed contrast enhancement in lacrimal sac mucocele walls[5]. In addition, dacryocyst diameters in these three planes after being measured in MRI images, 6.83-12.39 (mean 9.05) mm were observed in the horizontal length, 7.06-15.12 (mean 9.66) mm were observed in the sagittal length, and 8.64-16.01 (mean 11.43) mm were observed in the vertical length. To our knowledge, in adult, the sac generally measures 4-8 mm anteroposteriorly, and 3-5 mm horizontally[12-14]. Previous studies have indicated that patients with small lacrimal sac might have less effective rate of En-DCR than those with larger lacrimal sac and small lacrimal sac was considered to be a relative contraindication for DCR[15-19]. In this study, the lacrimal sac diameters of lacrimal sac mucocele was significantly larger than that of the normal lacrimal sacs. Big lacrimal sac has the following clinical significance. First, a large bony ostium should be considered during the surgery to completely expose the sac. Otherwise, if the bone is inadequately removed or the lacrimal sac (especially the lower end of the sac) is unable to open completely, the patient might present with postoperative purulent discharge while lacrimal irrigation was easily passed, which has been reported as the lacrimal sump syndrome. Infection caused by residual dacryocyst could also be the reason for the syndrome[20-21]. Second, a large posterior lacrimal flap can be made during the surgery, thereafter the sac flap can be stretched to the nasal mucosa with end-to-end anastomosing. These procedures might markedly improve the postoperative success rate[8].

Traditionally, a knife has been used to release purulent material from the lacrimal sac after probing[7,10]. In this study also, knife was used to incise the inferior anterior part of the sac to release the purulent material. Thereafter the lacrimal dust was conformed to patency by syringing through the inferior punctum. Finally, the sac was completely opened by the knife guided by a lacrimal probe and inserted through the superior punctum. These modifications were made, as it is impossible to cannulate in the patients with lacrimal sac mucocele. However, cannulation can be performed easily if the lacrimal sac is decompressed. Furthermore, the mechanism of cyst formation has suggested that a valve might exist at the common canalicular entrance[2-3,6]. Therefore, releasing the purulent material from the abscess to reduce the tension of the sac before implanting a probe could effectively prevent damage caused to the valve.

The need of silicone tube intubation in DCR surgery is controversial, as silicone tube is an inorganic material and therefore could cause intranasal tissue granulation formation, canalicular laceration and postoperative infection. Hence, such intubation is generally only recommended in the patients with small lacrimal sac, narrow upper nasal cavity, or canalicular stenosis/obstruction[22-27]. Silicone tubes were not used in the patients in the present study other than in 7 eyes with canalicular stenosis. The tubes were removed at 3mo postoperatively in an outpatient examination room.

Anatomical and functional success was achieved in 6 of the 7 intubation patients, with a success rate of 85.71%. As described above, the higher functional failure might be related to the dysfunction of the common canalicular, and intubation remains the main treatment method for the functional epiphora[28-30]. For the 17 functional failure patients in the study, tubes intubation was recommended, and 10 patients underwent the treatment. These tubes were then removed 2mo after intubation, and functional success was achieved in 8 of them. However, whether the implantation of silicone tube in En-DCR surgery can markedly improve postoperative functional success is another topic worth further studying.

The mass was found to be significantly shrunken when the lacrimal sac was opened during the surgery, and the swelling was completely relieved within 1-5d after the surgery, which concurred with the previously reported studies[1-2,4]. The swelling disappeared quickly probably because the mucocele was mainly caused by the fluid and mucus accumulation in the dacryocyst, and the infection was also inside the dacryocyst rather than causing inflammation beyond the dacryocyst.

Despite substantial efforts made to improve operative success rates, granulation tissue formation and scar tissue-mediated obstruction of the ostium were the primary causes of poor outcomes in the present study. For 9 cases with anatomical failure, failure occurred due to the scar formation at the ostium (n=5), granulation tissue formation at the ostium (n=3), and inferior canalicular obstruction (n=1). The prominence of ostium obstruction by scar or the granulation tissue as the main cause of poor operative outcomes was also consistent with prior studies[7,19,22].

In our series, surgical complications were relatively low. Electric coagulation was used to stop the bleeding in two patients when removing the bone. A cotton packing soaked in a vasoconstrictive solution was used to stop the postoperative epistaxis in a patient in the outpatient room. However, no severe complications such as prolapse of orbital fat, orbital hemorrhage, visual changes, or sinusitis were observed in this study.

There are multiple limitations associated with the present study. First, fluorescein staining was performed pre-operation in only a small subset of patients and was not randomly assigned according to the clinical findings, thus constraining the generalization of our results. Second, the optimal surgery time was not standardized, and hence additional research is required to confirm the value of timely En-DCR in lacrimal sac mucocele patients. Finally, more research is required to examine whether the silicone tube intubation at the time of En-DCR can significantly improve the functional success rate or not.

In conclusion, all the patients with ALSM had a history of NLDO. The interval between the obstruction and the onset of lacrimal sac mucocele varied. Mucoceles were found to be mild in the morning and severe in the evening in most of the cases. MRI examination has unique diagnostic characteristics for lacrimal sac mucocele and can be effectively used for the clinical diagnosis. An En-DCR is a good choice with a high anatomical success rate for the management of the patients with ALSM. However, the functional success rate was relatively low. Additional studies are required to confirm the value of timely En-DCR and the silicone tube intubation at the time of En-DCR in lacrimal sac mucocele.

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