14 Lacrimal Drainage System in Rhino-Orbito-Cerebral Mucormycosis

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A unique chapter on lacrimal drainage system (LDS) in rhino-orbito-cerebral mucormycosis (ROCM) is included to highlight the various changes in LDS (especially the lacrimal sac and nasolacrimal duct) seen during surgical management of ROCM in covid pandemic. Never before have such a large number of sac and nasolacrimal ducts been investigated for the amount of their involvement in mucormycosis as well as their microbiological and histological features.

Endoscopic Cleaning of pterygopalatine fossa (PPF) needs endoscopic median maxillectomy for surgical care of ROCM. In all the cases where medial wall of maxilla is removed, the nasolacrimal duct (NLD) is exposed. The clinical presentation of mucormycosis involving LDS, imaging, diagnostic confirmation, surgical methods involving the handling of LDS, and LDS involvement in aspergillosis in comparison to mucormycosis are all discussed in this chapter.

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14.1 Various Appearances of the sac and NLD

The steps for exposing the sac and NLD are the same as those outlined in the chapter on surgical management of ROCM. All of the cases underwent drilling of the bone covering the sac and NLD after a control hole was drilled in anterolateral wall of maxilla during Denker's modified medial maxillectomy. In cases with disease limited to the pterygopalatine fossa, the NLD wall may not be impacted (Figs. 14.1, 14.2, and 14.3). NLD is traced upto the lacrimal sac and the lower part of the sac is also examined. For the complete clearance of disease from pterygopalatine fossa, NLD needs to be severed. In an oblique fashion, it is sliced flush with the orbital floor. The sac is left in place, and the oblique cut creates a larger lumen in the sac, allowing for tear drainage. The resected NLD is examined (Figs. 14.4, 14.5, and 14.6) and while resecting no part of NLD should be left hanging in the nasal cavity. Any remaining NLD left hanging into the nose undergoes fibrosis, closure and causes epiphora.

In the case of inflammatory tissue and mucopurulent discharge filling the antrum, the NLD wall was involved with bluish discolouration and thick wall (Fig. 14.7). After removing the inferior turbinate (yellow arrow) the distal end of NLD was examined using a probe in situ (black arrow) (Fig. 14.8). The NLD was split vertically, and the inspissated purulent discharge and inflammatory

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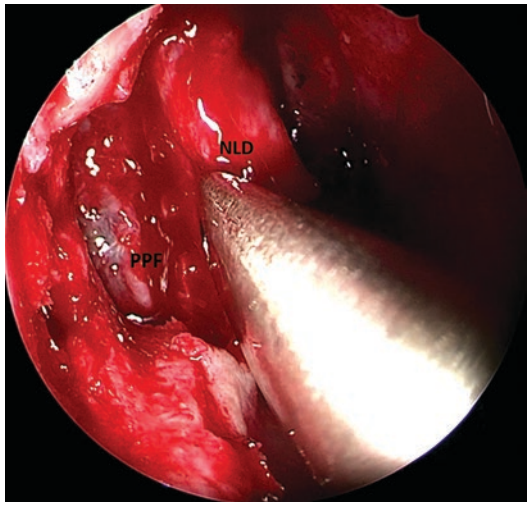

Fig. 14.1 Endoscopic view of the right nasal cavity demonstrating a thin-walled nasolacrimal duct (NLD) with the disease in the right maxillary antrum. (*PPF* Pterygopalatine fossa)

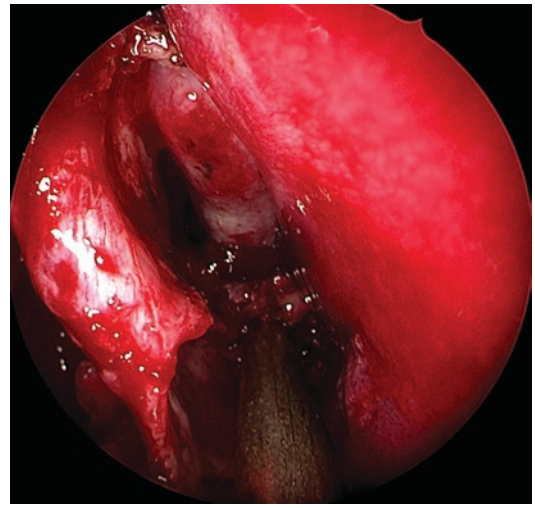

Fig. 14.3 A view of the lower part of the right sac and NLD from a close distance

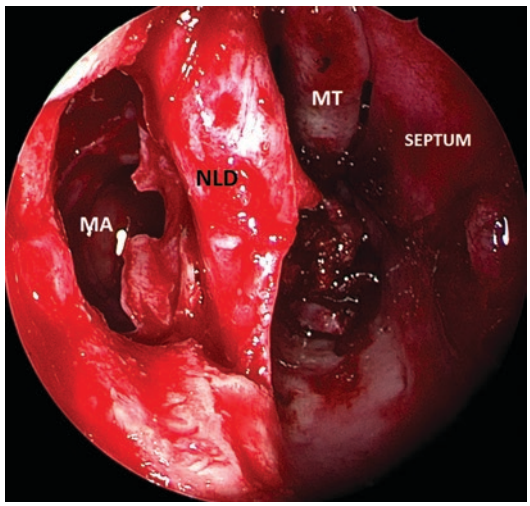

Fig. 14.2 A pinkish lower part of the sac and NLD with normal walls in a case of limited disease. (*MA* Maxillary antrum, *MT* Middle turbinate)

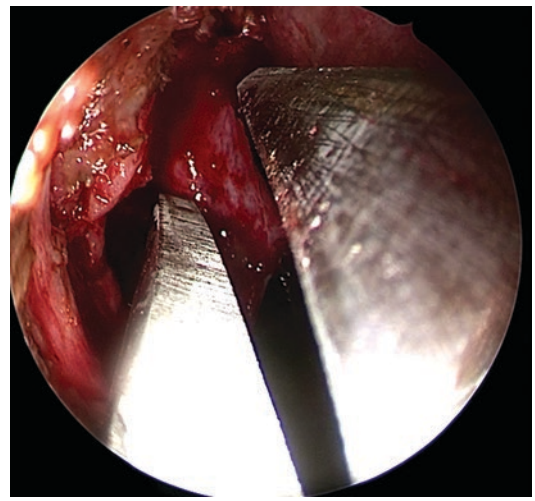

Fig. 14.4 A pair of scissors is inserted at the sac duct junction

tissue were removed from the antrum (Figs. 14.9 and 14.10). The NLD lumen was found to be patent with only the inflammatory alterations in the wall (Fig. 14.11). The NLD was cut out and analysed (Fig. 14.12).

In cases of extensive disease filling the nose, maxillary sinus, pterygopalatine fossa, the NLD was thickened, pale and thrombotic (Fig. 14.13). In such cases, the frontal process of maxilla is drilled higher up until the orbital floor so that no bone is left over the sac duct junction (Fig. 14.14). It allows NLD severance more proximal with no postoperative epiphora (Fig. 14.15).

In cases where the lower part of the sac is seen on proximal drilling of the bone that looks healthy (Fig. 14.16), it can be marsupialized at

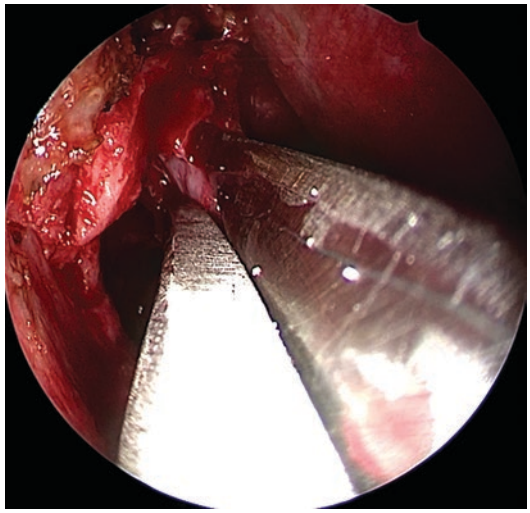

Fig. 14.5 The NLD is incised at sac duct junction and flushed to the orbital floor so that it does not hang into the nose

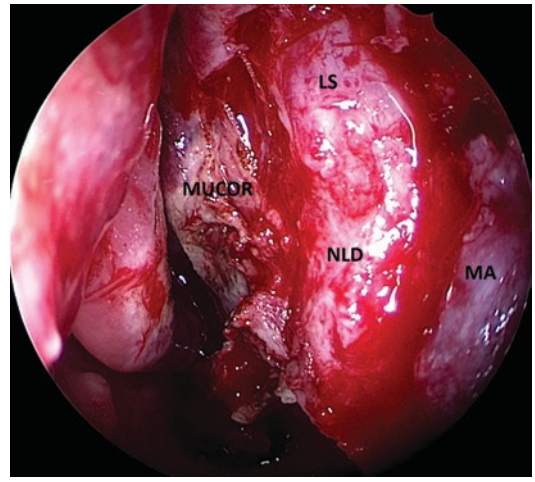

Fig. 14.7 An oedematous bluish and inflamed lacrimal sac and NLD appears to be surrounded by disease filled maxillary. (*MA* Maxillary antrum, *Ls* Lacrimal sac, *NLD* Nasolacrimal duct)

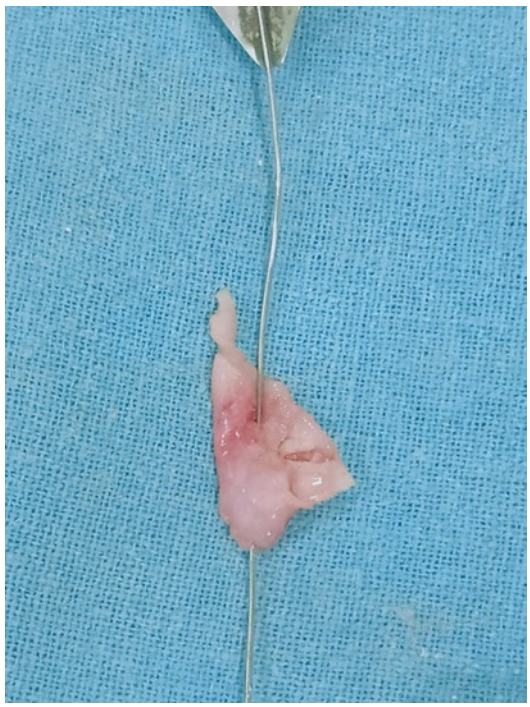

Fig. 14.6 The removed NLD is pinkish in colour and appears to be free of inflammation

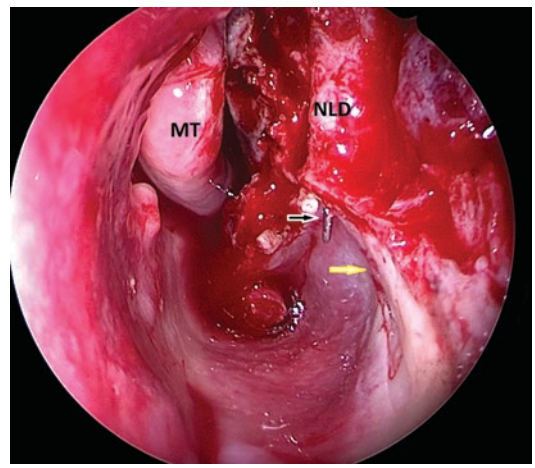

Fig. 14.8 The inferior meatus (yellow arrow) with a probe in situ (white arrow) in the distal opening of NLD. (*MT* Middle turbinate)

this level. In other cases, the bone of the frontal process of the maxilla may look unhealthy. The underlying sac and NLD are often thrombosed and may need a more superior resection (Figs. 14.17 and 14.18).

The full NLD appearance in cases of significant orbital disease may vary from a damaged friable and inflamed NLD to a whitish necrotic rope-like structure (Figs. 14.19 and 14.20). In a widespread disease a completely melted NLD remnant may be visible (Fig. 14.21).

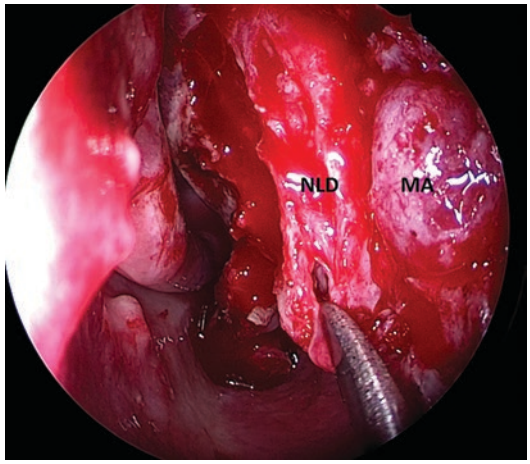

Fig. 14.9 The NLD is split open vertically (*MA* Maxillary antrum)

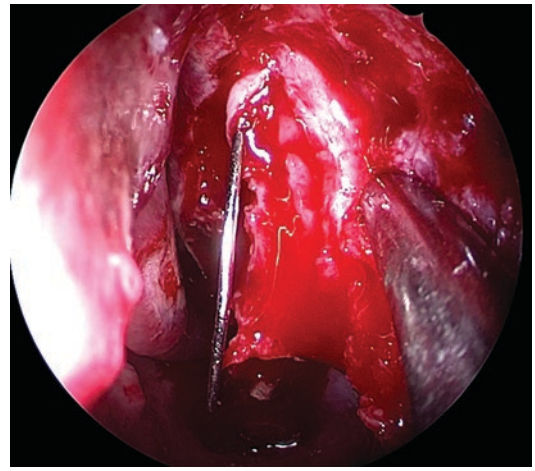

Fig. 14.11 Antrum is cleared, and the probe is rotated in all the directions to examine the NLD lumen

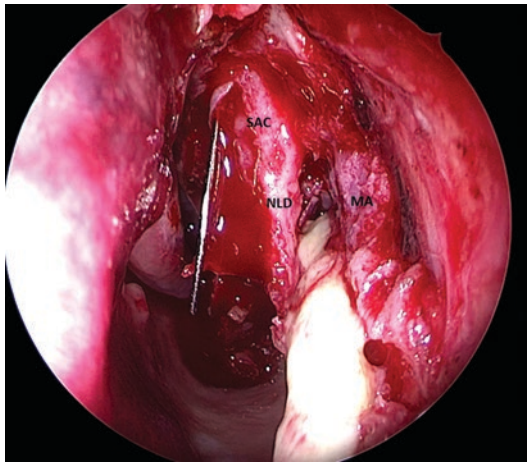

Fig. 14.10 The lumen of NLD seems clean, but the purulent discharge can be observed flowing out of the maxillary antrum (*MA* Maxillary antrum)

Fig. 14.12 Excised NLD looks inflamed and oedematous

14.2 Clinical Presentation of Mucormycosis Involving LDS

Patients with LDS involvement in ROCM presented with a variety of clinical symptoms, which varied depending on the severity of the cases. Epiphora, necrosis of the skin around the medial canthal (Fig. 14.22), and a large fistula involving the punta and canaliculi were among the symptoms.

14.3 Imaging

CT scans and MRI revealed involvement of the LDS as a component of other sinonasal structures. LDS lesions were seen as a hypodense mass with evidence of soft tissue density lesion on coronal CT section in the left lacrimal sac area with distorted bony NLD, compared to a normal

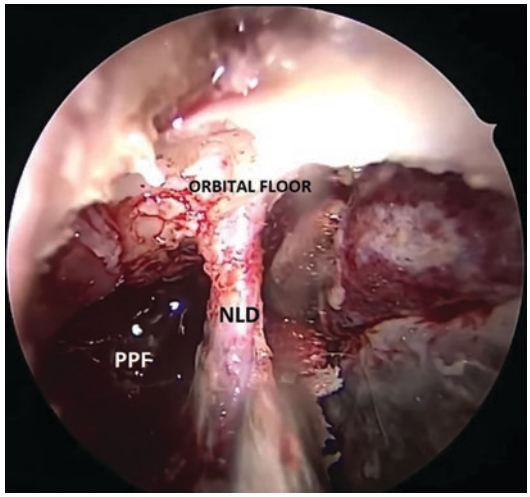

Fig. 14.13 In a case of disease filling the antrum, the NLD looks pale and unhealthy. (*PPF* Pterygopalatine fossa) (Photo courtesy: Dr Satish Jain Jaipur)

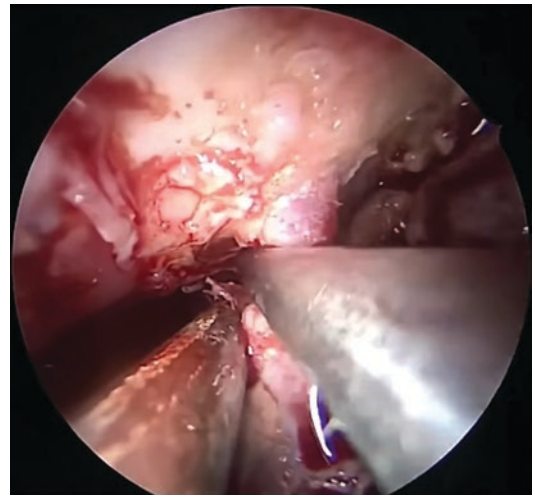

Fig. 14.15 The NLD is cut flush with the orbital floor (Photo courtesy: Dr Satish Jain Jaipur)

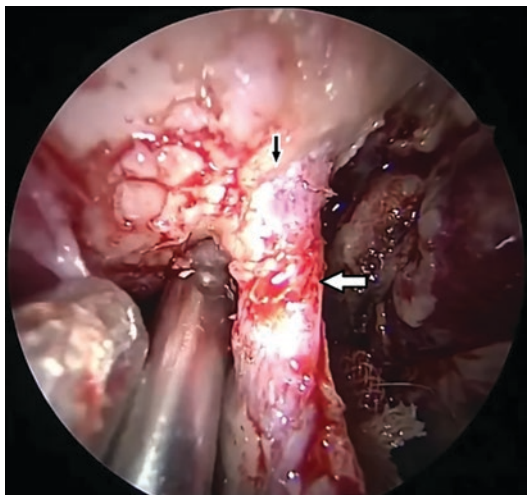

Fig. 14.14 The overlying bone is drilled all the way to the sac duct junction (white arrow), and then the bone is drilled till the orbital floor (white arrow) (Photo courtesy: Dr Satish Jain Jaipur)

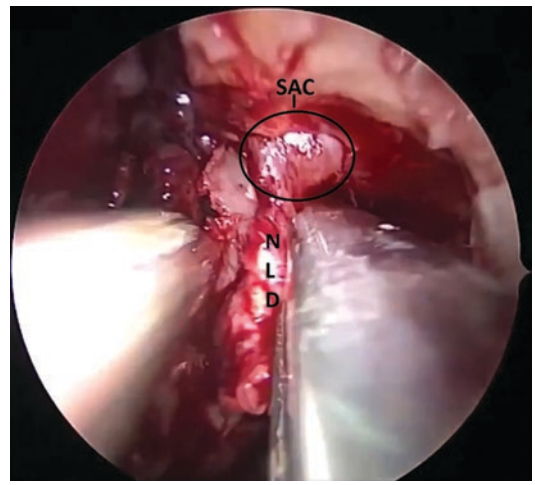

Fig. 14.16 The level of a marsupialization of the lacrimal drainage system is determined by the sac's state. The lower section of the sac is exposed in this case the sac, and it appears to be healthy (black circle). A sharp incision is given to cut the NLD obliquely (Photo courtesy: Dr Satish Jain Jaipur)

nasolacrimal system on the right side (Figs. 14.23 and 14.24). On axial sections, the disease was seen in the left maxillary antrum reaching up to the NLD and lamina papyracea (Fig. 14.25). On gadolinium-enhanced MRI, different zones of enhancement and non-enhancement were detected in the lacrimal sac and surrounding ethmoid cells in an extensive case of ROCM (Fig. 14.26).

14.4 Diagnostic Confirmation

14.4.1 Microscopy

The material is inoculated on Sabouraud dextrose agar and brain heart infusion agar [1] (Fig. 14.27).

Direct microscopy allows for a rapid diagnosis based on septation, angle of branching and

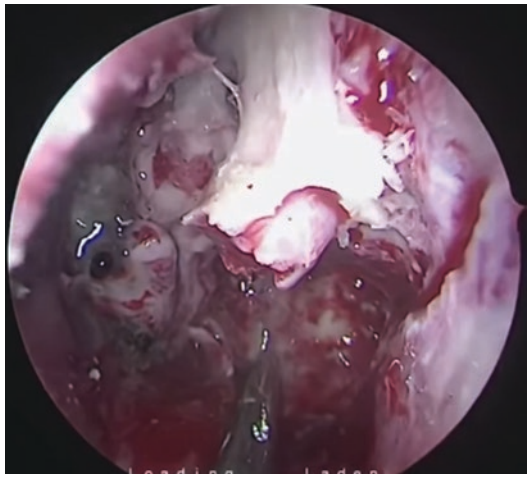

Fig. 14.17 In cases of extensive disease filling the pterygopalatine fossa, the frontal process of the maxilla is generally unhealthy. On drilling it, a totally thrombosed NLD can be noticed (Photo courtesy: Dr Satish Jain Jaipur)

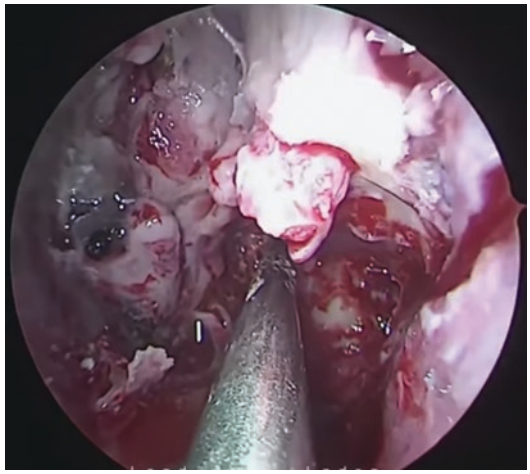

Fig. 14.18 When probing the cut end no lumen could be found

width of the hyphae. Potassium hydroxide (KOH) mount in 10–20% KOH preparation reveals characteristic broad, ribbon-like aseptate hyphae of Mucorales on direct microscopy. Under a fluorescent microscope KOH-Calcofluor white solution enhances the visualization of the fungal element in specimens.

14.4.2 Histopathology

Histopathology revealed fungal hyphae with a ribbon-like appearance with random branching at various angles (Fig. 14.28).

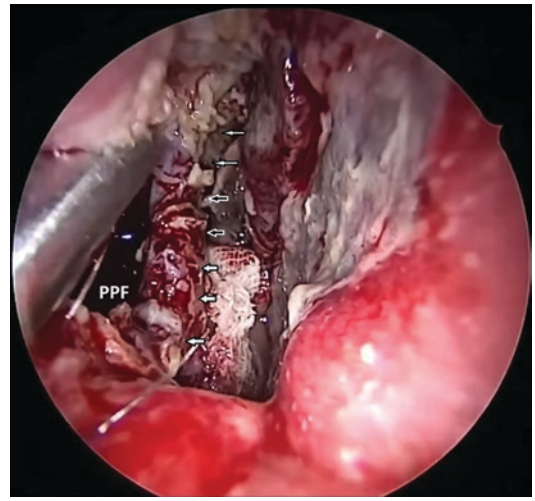

Fig. 14.19 In an instance of significant disease a friable necrotic and sloughed-out right NLD was seen (arrows) (*PPF* Pterygopalatine fossa) (Photo courtesy: Dr Satish Jain Jaipur)

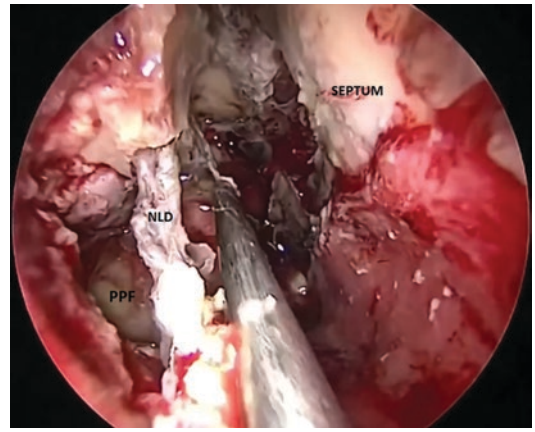

Fig. 14.20 A completely necrotic thinned-out rope-like NLD with slough and blackish discolouration (*PPF* Pterygopalatine fossa) (Photo courtesy: Dr Satish Jain Jaipur)

14.5 NLD as a Possible Disease Spread Route to the Orbit

NLD has been mentioned by several publications as a probable route of spread of disease transmission from the nose to orbit [2–7].

According to them, the orbital involvement is caused by nasolacrimal duct invasion which then spreads to the orbit via the thin medial orbital wall [2]. However, there was no specific evidence for us to prove that. The inferior

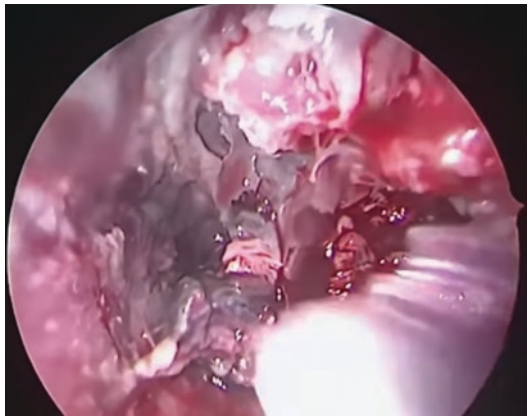

Fig. 14.21 A melted and damaged NLD (Photo courtesy: Dr Satish Jain Jaipur)

Fig. 14.22 External photograph of a patient with mucormycosis of the lacrimal drainage system (LDS), showing the diffuse involvement of the puncta and canaliculi secondary to the involvement of sinuses

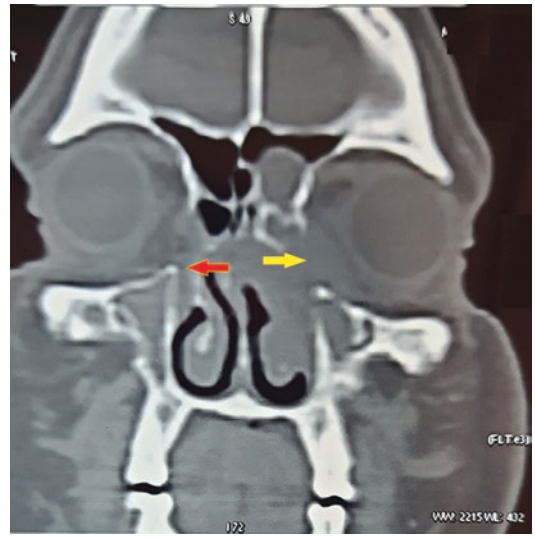

Fig. 14.23 Computed tomographic scan of the patient demonstrating soft tissue density in the left maxillary antrum and ethmoid cells along with the involvement of left LDS (yellow arrow) and a normal LDS on the right (red arrow)

Fig. 14.24 Computed tomographic scan coronal section showing soft tissue density filling the left-sided sinuses with involvement of the lacrimal drainage system

meatus was normal, and in the cases where NLD was patent, the medial canthal area was pressed, and the discharge could be seen accumulating at the distal end of NLD, indicating a patent NLD (Fig. 14.29). A sample from this location was sent to the lab for fungal culture (Fig. 14.30), which was found to be mucor free. Patients, however, did have orbital disease indicating a different route of dissemination.

In cases of widespread disease, NLD itself was engulfed and eroded by the disease that surrounded it from all sides. In advanced cases, the orbital disease was massive, pterygopalatine fossa was filled with the disease and NLD had secondary involvement. It suggests that the disease did not spread to the orbit through NLD. There was no instance in which NLD involvement and infiltration with the mucormycosis was greater than the surrounding disease, and vice versa. It appears that NLD involvement was secondary in all of these cases.

There has never been a comprehensive examination of the extent and kind of LDS involvement as well as emergence and therapy in invasive fungal infections.

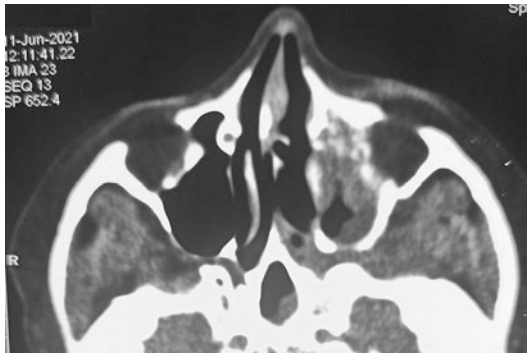

Fig. 14.25 Erosion of the left bony NLD with opacified maxillary antrum

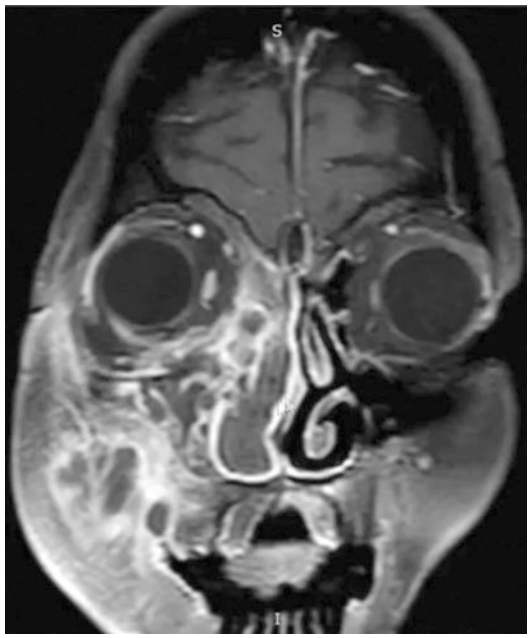

Fig. 14.26 MRI showing diverse areas of enhancement and non-enhancement in the right lacrimal sac is in a case with widespread disease

14.6 Surgical Procedures Involving LDS

LDS is a vital structure that we come across during the management of various sinonasal conditions like inverted papilloma, juvenile nasopharyngeal angiofibroma, sinonasal tumours and invasive fungal disorders of the sino-orbital areas. The approaches involving LDS are Denker's modified medial maxillectomy in which

Fig. 14.27 Fungal growth was seen on Sabouraud agar and brain heart infusion agar

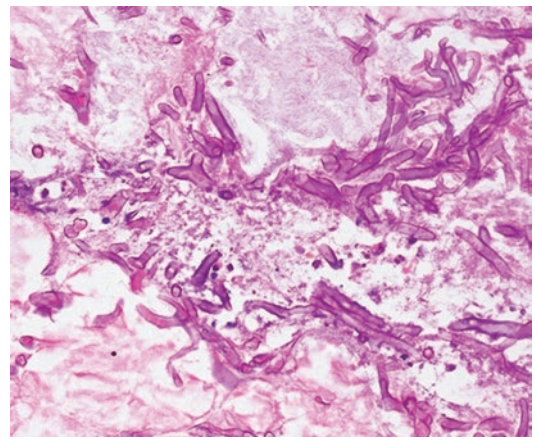

Fig. 14.28 Fungal hyphae with a ribbon-like appearance with random branching at variable angles (Photo courtesy; Dr Aanchal Kakkar, AIIMS Delhi)

a window is created first and then distal half of the sac and the nasolacrimal duct are exposed

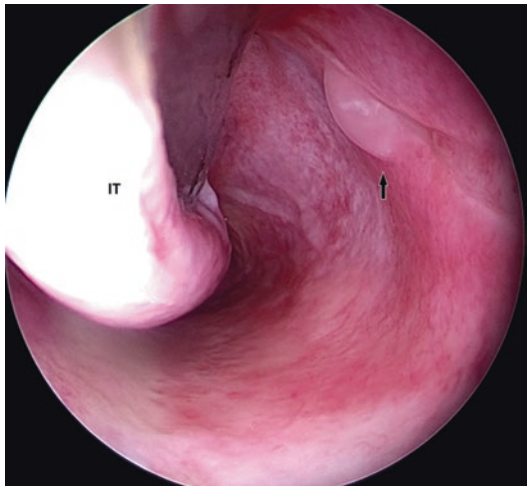

Fig. 14.29 Discharge can be seen flowing into the nose through the NLD opening into the inferior meatus (arrow), on pressing over the medial canthus. IT; Inferior turbinate

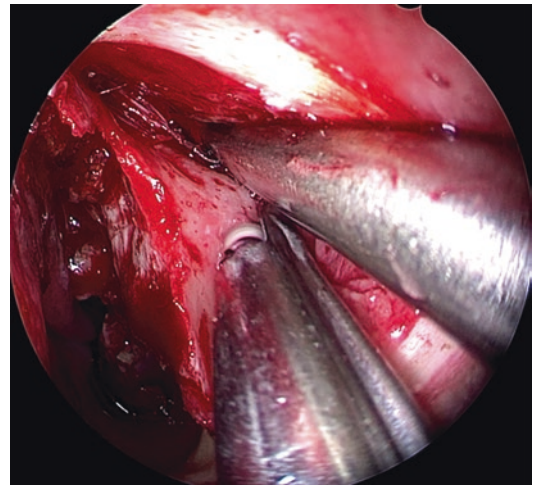

Fig. 14.31 Drilling a hole into the anterolateral wall of the maxilla for the inspection of disease inside the antrum during Denker's medial maxillectomy

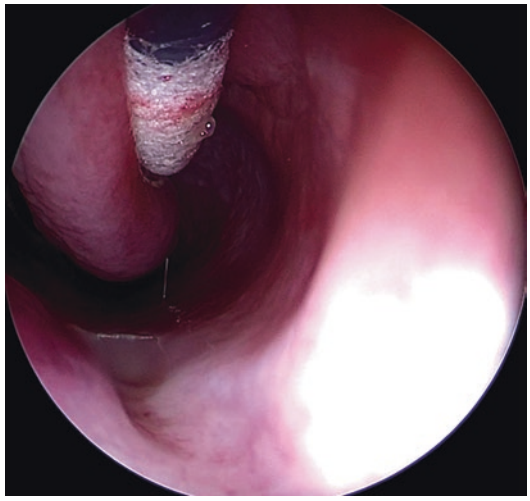

Fig. 14.30 A swab from the inferior meatus is obtained and sent for culture

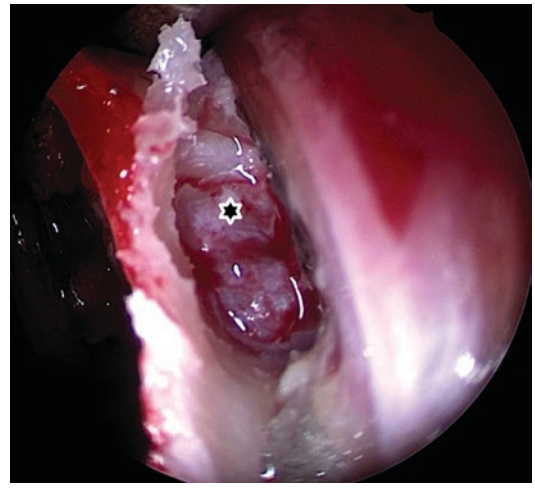

Fig. 14.32 Maxillary antrum has been opened up and can be seen filled with the disease (star). The medial wall of the maxilla overlying the NLD is still intact

(Figs. 14.31, 14.32, and 14.33). In Denker's NLD is severed to access the disease in the pterygopalatine fossa while in prelacrimal approach NLD may be preserved.

Denker's modified medial maxillectomy approach is the most popular approach for accessing the disease in the maxillary sinus, pterygopalatine fossa and infratemporal fossa [8–11]. The classic medial maxillectomy involves the resection of the whole medial maxillary sinus wall, including the inferior turbinate and the nasolacrimal duct [12–14].

The prelacrimal approach is used to gain improved access by resecting a part of the piriform aperture and the anterior wall of the maxillary sinus. In this approach, inferior turbinate is temporarily displaced to gain access and then repositioned to reduce morbidity. This procedure provides excellent visualization of the ante-

rior wall of the maxillary sinus and the prelacrimal recess. It allows the inferior turbinate and the nasolacrimal duct to be preserved [15–28].

14.6.1 Medical Management

Liposomal amphotericin B is the medication of choice for mucormycosis. The chapter on antifungal treatment has more information on the doses and types of amphotericin B that are available.

There are only a few cases of primary mucormycosis of the lacrimal sac in the literature. One of them is of a 72-year-old woman with several dacryoliths and mucormycosis on histopathology [29]. Another patient was a known case of diabetic ketoacidosis who was operated for lacrimal sac infection, and on histopathology revealed mucormycosis [30].

Let us look at the other type of invasive fungal infection of the lacrimal sac, aspergillosis, which was seen during the Covid epidemic.

14.7 Aspergillosis of the LDS

Aspergillosis is a slower-progressing lesion of the LDS as compared to mucormycosis and is also less deadly. Owing to slow progression, sometimes patients present with lacrimal sac abscesses, and the biopsy reveals aspergillosis. A case of aspergillosis masquerading as lacrimal sac abscess has been reported [31].

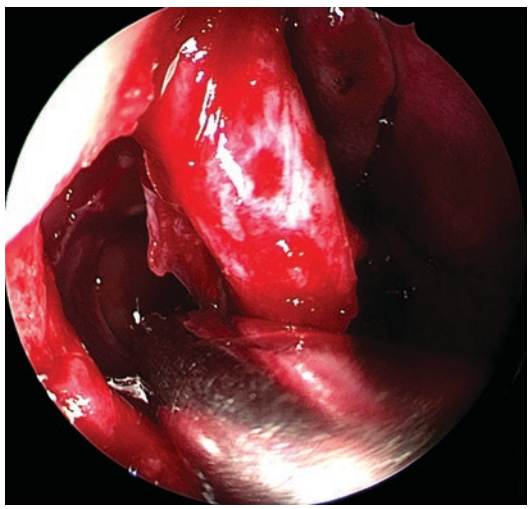

Fig. 14.33 Premaxillary approach: the NLD (right side in this case) can be kept intact depending on the indication

14.7.1 Clinical Presentation

The presenting symptom in invasive aspergillosis of LDS is epiphora with medial canthal swelling. However, as previously mentioned, it can present as a lacrimal sac abscess, which can cause confusion for the surgeon, and if a biopsy is not performed during surgery, the diagnosis may be missed.

Epiphora is generally the first sign followed by swelling that develops after a long gap. It shows that NLD is the earliest site of aspergillosis inoculation, as its narrow lumen causes it to get clogged early. Further disease progression develops a mass lesion in the lacrimal sac, which starts distending and manifests as a swelling in the medial canthal region.

14.7.2 Examination

External examination shows a mass in the medial canthal area extending laterally along the inferior lid margin (Fig. 14.34). Nasal endoscopy shows a bulge over the lateral wall encroaching the premaxillary line with prominent blood vessels across it (Fig. 14.35).

14.7.3 Imaging in Aspergillo

A significant soft tissue mass in the right lacrimal fossa, a breach in the lacrimal bone and

Fig. 14.34 External photograph of a patient with invasive aspergillosis of the right lacrimal drainage system

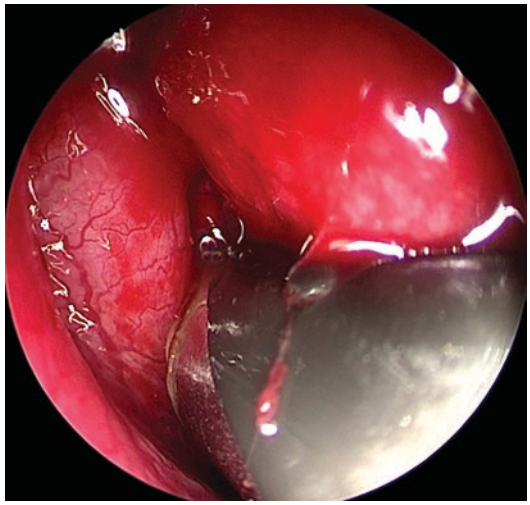

Fig. 14.35 An endoscopic view of the right nasal cavity with a protrusion in the nasolacrimal area

lamina papyracea with soft tissue extension into the ethmoid sinuses can be seen on computed tomography (CT) scan coronal (Fig. 14.36), axial (Fig. 14.37) and sagittal sections (Fig. 14.38) in aspergillosis of the lacrimal sac. An extension of the soft tissue oedema into the inferior meatus is seen.

14.7.4 Management of Aspergillosis of the Lacrimal Sac

In aspergillosis, unlike LDS mucormycosis, the whole sac and NLD are filled with a solid mass. Coblation of the prominent vessels over the bulging lateral wall is one of the measures in treating LDS aspergillosis (Fig. 14.39). After that a blunt periosteal elevator is used to make an incision. The frontal process of maxilla is seen after the dissected portion is lifted and removed (Fig. 14.40).

The bone of the frontal process of the maxilla is removed using a Kerrison punch (Fig. 14.41). The lacrimal sac is opened, and the mass is examined. Aspergillus is a firm mass and has rubber-like consistency (Fig. 14.42). It is often peeled off from the underlying structures and is debrided. The superior part of the sac is palpated (Fig. 14.43). The lumen of the sac is examined,

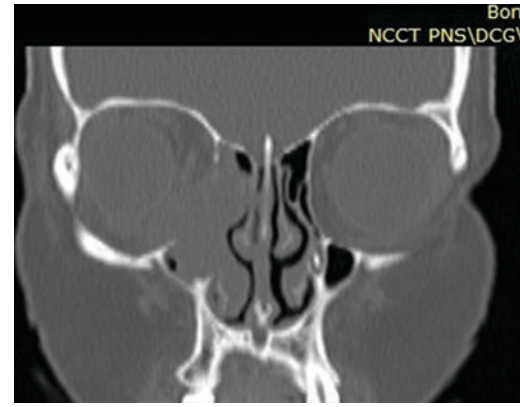

Fig. 14.36 Non-contrast computed tomography (NCCT) of sinuses and orbit, coronal section demonstrating soft tissue mass occupying the right lacrimal sac, NLD and ethmoids

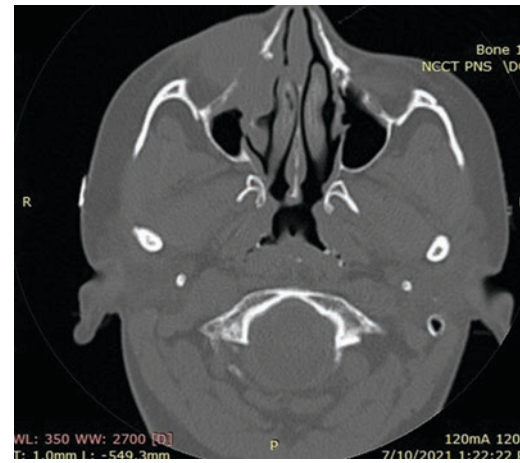

Fig. 14.37 NCCT of sinuses and orbit axial section demonstrating soft tissue mass occupying the right lacrimal sac, NLD and ethmoids

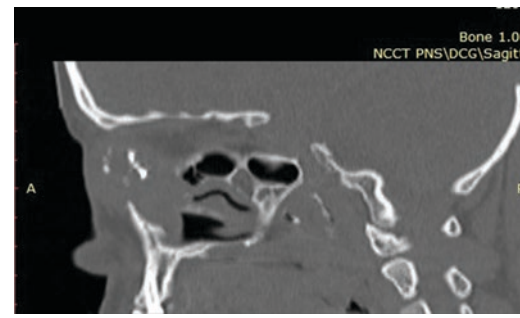

Fig. 14.38 Sagittal section on CT DCG showing a mass filling the lacrimal sac, NLD and the surrounding area

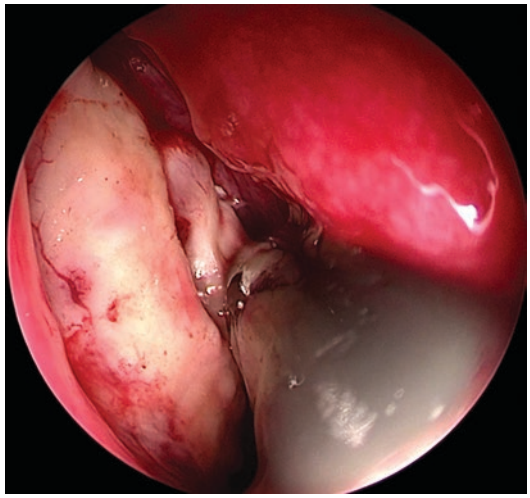

Fig. 14.39 The lateral wall bulge is coblated to minimize bleeding

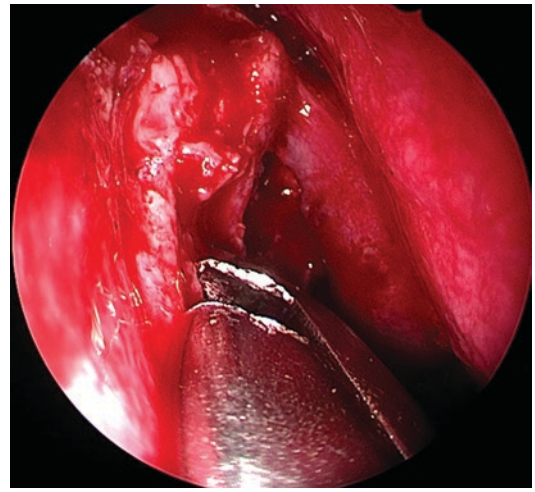

Fig. 14.41 Kerrison punch is used to remove the bone of the frontal process of the maxilla

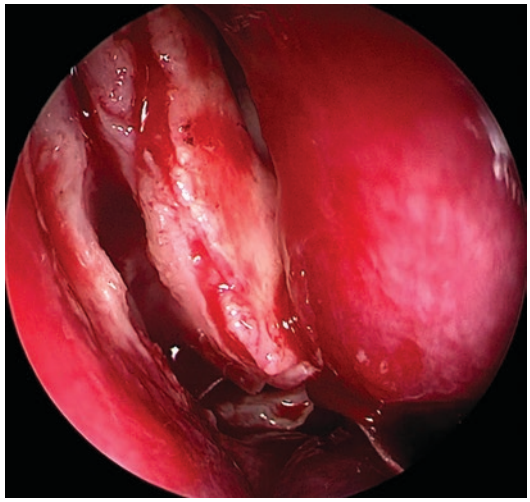

Fig. 14.40 The hypertrophied tissue on the lateral wall is lifted off in continuation with the uncinate process

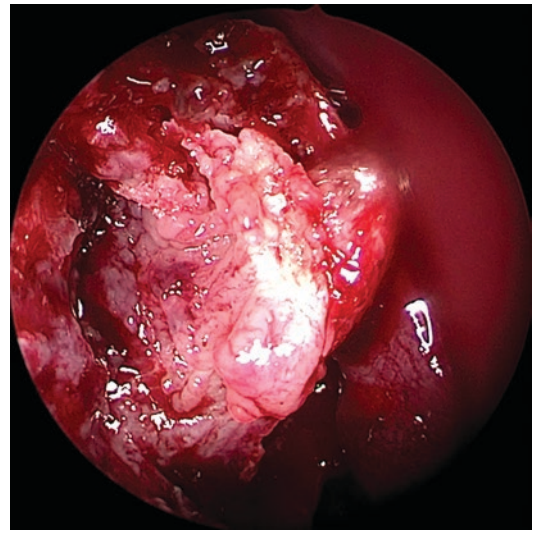

Fig. 14.42 The rubber-like mass is seen filling the sac, occluding its lumen. The tissue filling the sac is teased out and debrided

and the probe can be seen in situ (Figs. 14.44 and 14.45). After clearing the fungal mass, the sac and NLD are then dissected off the fossa, a dacryocystectomy is carried out, and the specimen is sent to the lab.

14.7.5 Microbiology and Histopathology

KOH mount showed bright green septate hyphae of *aspergillosis* under *a* fluorescence microscope (Fig. 14.46). Microphotograph shows PAS-stained giant cells along with septate hyphae in the granulomatous area (Fig. 14.47).

14.7.6 Antifungals and Prognosis

Aspergillosis responds to voriconazole that is available in intravenous as well as oral formulations and is well tolerated. We found only a

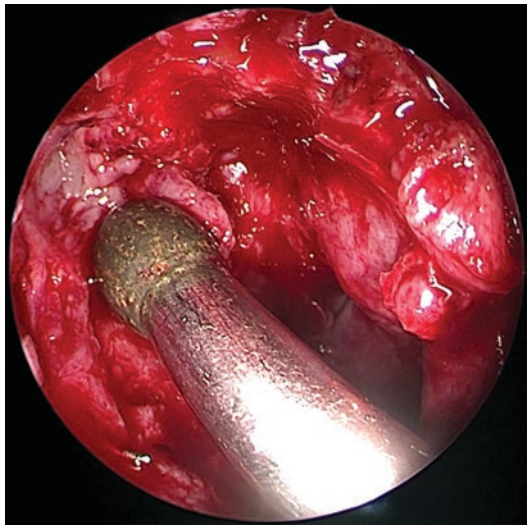

Fig. 14.43 The superior part of the sac is checked with a curved blunt suction tip, and any residual disease is debrided

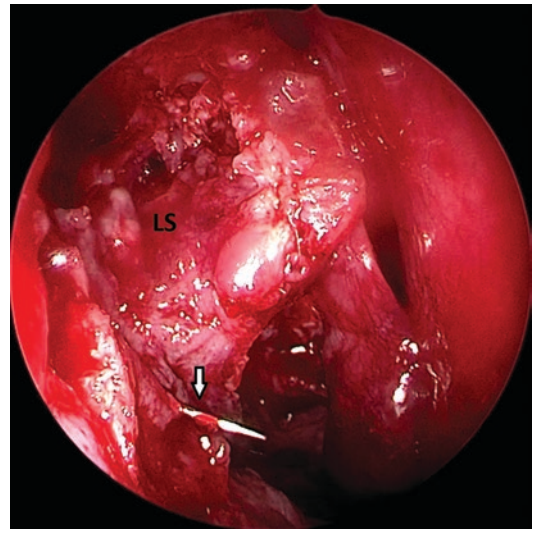

Fig. 14.45 Probe can be seen in situ (arrow). (*LS* lacrimal sac)

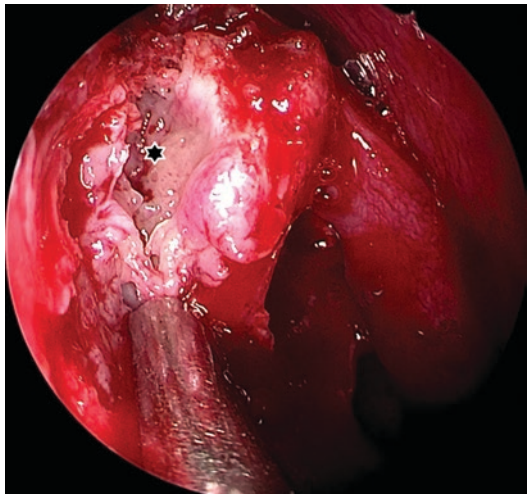

Fig. 14.44 Sac lumen is visible after disease clearance (star). The soft tissue mass seen all around the lumen is debrided

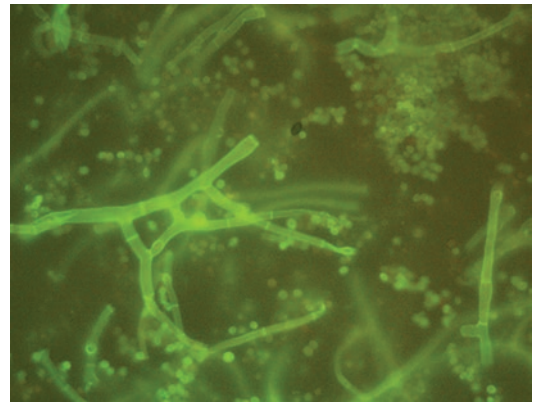

Fig. 14.46 KOH mount showing bright green septate hyphae of aspergillosis under fluorescence microscope (Photo courtesy: Dr Arpan Gandhi, SCEH, Delhi)

few cases of isolated lacrimal sac aspergillosis in literature [31, 32]. The cases were successfully treated with surgery and antifungal medication.

Invasive fungal infections of the isolated lacrimal drainage system are rare. Very few cases of invasive mucormycosis and aspergillosis of the lacrimal drainage system (LDS) have been reported in the literature [29–32]. The second wave of Covid pandemic in India resulted in an unexpected increase in invasive fungal infections of LDS. Although incidences of LDS mucormycosis were discovered during an endemic of rhino-orbito-cerebral (ROC) mucormycosis, occurrences of LDS aspergillosis were also on the rise.

14.8 Comparison of LDS Aspergillosis and LDS Mucormycosis

The features of various invasive fungal infections of the lacrimal drainage system are compared in Table 14.1.

Prognosis of invasive fungal infection of LDS is good with surgery, antifungal drugs and the management of underlying medical conditions. To summarise, in aspergillosis, ROCM surgical debridement entails sacrificing the NLD which is cut flush to the floor of the orbit, and performing a dacryocystectomy with complete sac and NLD removal.

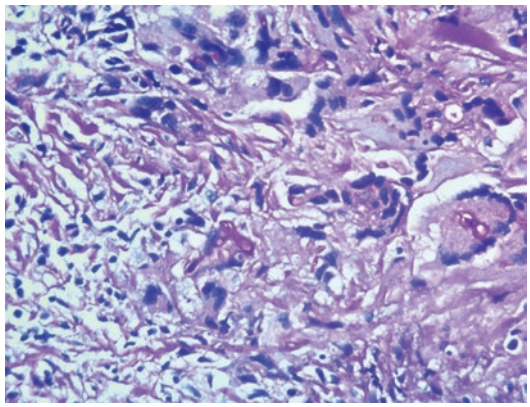

Fig. 14.47 Microphotograph showing PAS-stained giant cells along with septate hyphae in the granulomatous area—20× (Photo courtesy: Dr Arpan Gandhi, SCEH, Delhi)

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Table 14.1 Invasive fungal infection of the lacrimal drainage system: comparison of aspergillosis and mucormycosis

SN	Clinical features	Aspergillosis	Mucormycosis
1	Swelling in the medial canthal area	Present	May or may not be present
2	Onset	Slow, may get detected only after secondary infection sets in	Incidental finding [1, 2] or seen in association with the sinonasal disease with rapid progression during Covid
3	Isolated LDS involvement	Has been seen [3, 8]	Not seen in our cases, though reported earlier [1, 2]
4	Management	Surgery and voriconazole	Surgery and amphotericin B
5	Intraoperative	Consistency is rubber like and difficult to debride	Soft and easy to debride
6	Histopathology	Septate hyphae	Aseptate hyphae

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