

ISOLATED AGGER NASI MUCOCOELE: A FACTOR FOR RECURRENCE?

Sangeetha Thirumazhisi Sachithanandam¹

¹Senior Registrar, Department of ENT, Global Hospitals, Chennai.

ABSTRACT

Paranasal sinus mucocoeles are more common in frontal and ethmoidal sinuses with highest morbidity and recurrence rates. Still, endoscopic surgical clearance of disease in the frontal recess area is technically challenging in view of recurrence. A case of recurrent isolated agger nasi mucocoele is reported here and a detail analysis is made on the anatomical, disease and surgical factors for the recurrence of mucocoeles in frontal recess area, the radiological aspects of such lesions required for an operating ENT surgeon and the best surgical approaches to the frontal recess area. Axillary flap technique designed by Wormald is adopted for the surgical clearance of this recurrent isolated agger nasi mucocoele and is found to be safe, easy with excellent anterior access to the frontal recess area preventing postoperative adhesions, scarring and recurrence.

KEYWORDS

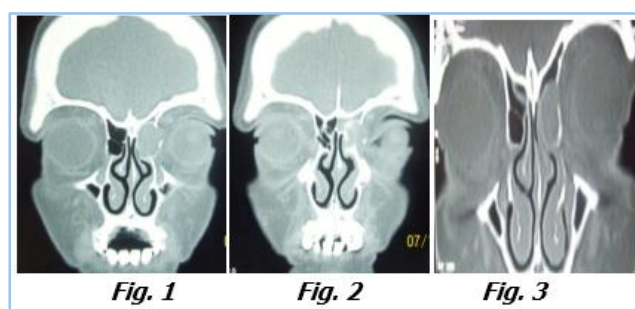
Agger Nasi Mucocoele, Recurrence, Best Approach to Frontal Recess, Axillary Flap Technique.

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INTRODUCTION: Paranasal sinus mucocoeles are benign slow growing mucus retention cysts creating a varied presentation extending from a small intranasal cyst to a large expansile osteolytic lesion with intracranial and intra-orbital extension. Frontoethmoidal region is the most common site for occurrence as well as recurrence of the mucocoeles with high morbidity. A case of recurrent isolated agger nasi mucocoele is reported here and an attempt is made to analyse the critical anatomical, disease and surgical factors for the recurrence of mucocoeles in frontal recess area and the best surgical approaches to frontal recess to avoid future recurrences. Axillary flap technique designed by Wormald is adopted for the surgical clearance of this recurrent isolated agger nasi mucocoele and is found to be safe, easy with excellent anterior access to the frontal recess area preventing postoperative adhesions, scarring and recurrence.

Case Details: A 42-year-old female presented to the ENT outpatient department with the complaints of pain in and around the left eye which is persistent dull aching, aggravated on eye ball rowing movements for 1 month. No history of epiphora, vision impairment or diplopia. Previous history of similar complaints was present before one year for which she consulted an ophthalmologist where her visual and lacrimal systems were reported as normal. She was referred to an ENT hospital and CT-nose and paranasal sinuses was advised to her. A single homogenous cystic lesion was seen involving the left agger nasi cell which was expansile and bony remodelling with thinning of lamina papyracea was present [Fig. 1, Fig. 2].

She underwent endoscopic sinus surgery with no further records on operative procedure and findings. Clinical examination of the patient revealed nil significant findings other than tenderness near the left eye medial canthus. Diagnostic nasal endoscopy showed a well-healed mucosa with clear anterior ethmoidectomy and wide middle meatal antrostomy in the middle meatus. A bulge is noted in the left agger nasi region. Repeat CT-paranasal sinuses was done which showed a homogeneous hypodense cystic lesion involving the left agger nasi cell? Mucocoele [Fig. 3]. Endoscopic marsupialisation of the recurrent mucocoele by axillary flap technique was done for this patient. This article is made to enlighten the critical anatomical and surgical factors for the recurrence of mucocoeles in the frontal recess area, the radiological requirements for an operating ENT surgeon in such cases and the best surgical approaches to frontal recess to avoid future recurrences.



DISCUSSION: Mucocoeles of the paranasal sinuses were first described by Langenbeck (1820) as hydatids. Rollet (1909) named them as mucocoeles.^[1] Paranasal sinus mucocoeles are mucus filled cystic masses due to the obstruction of sinus ostia. They are benign slow growing masses lined by respiratory epithelium with expansile and osteolytic properties due to mechanical compression and osteoclastic bone resorption induced by inflammatory mediators like cytokines (IL-1, IL-6), TNF-alpha, prostaglandins (PGE₂) and collagenases producing reduced vascularisation of bone.^[2,3]

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Corresponding Author:

Dr. Sangeetha Thirumazhisi Sachithanandam,

No: 56/96, T. S. V. Koil Street, Mylapore,

Chennai-600004.

E-mail: drsangavy@gmail.com

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Frontal sinus and ethmoid sinus mucocoeles are more common (70-90%) compared to maxillary sinus (10%) and sphenoid sinus mucocoeles which are rare.^[4] Any inflammatory obstruction to the sinus ostia or cystic dilatation of the mucosal glands or polypoidal degeneration of the lining epithelium are the primary causes for mucocoele formation. Other causes include previous sinus surgery, chronic sinusitis, facial trauma, sinonasal manifestations of systemic diseases like cystic fibrosis, fibrous dysplasia, osteoma or ossifying fibroma. Paranasal sinus mucocoeles are more common in 30-40 years of age with equal sex distribution. Varying symptoms are produced due to mass effect depending on the location of the mucocoele which are grouped into ophthalmological, rhinological and neurological symptoms. Frontoethmoidal mucocoeles commonly present with proptosis, diplopia, periorbital swelling due to mass effect upon the orbit.^[1] Frontoethmoidal region is the most common site for occurrence as well as recurrence of the mucocoeles.

In an attempt to analyse the factors for the recurrence of mucocoeles in frontal recess area, knowledge on the 3-dimensional anatomy of the frontal recess and its cells, acquires the prime importance. The boundaries of frontal recess or frontal sinus outflow tract are middle turbinate medially, lamina papyracea laterally, anterior skull base with anterior ethmoidal artery superiorly, anterior face of bulla posteriorly and anterior wall of agger nasi anteriorly. The frontal cells above the agger nasi cell in the frontal recess area are classified into 4 types using Kuhn classification.

Frontal Cells- Kuhn classification.

Type 1 Single frontal recess cell above agger nasi.

Type 2 Tier of cells in frontal recess above agger nasi.

Type 3 Single massive cell pneumatizing into frontal sinus.

Type 4 Isolated cell in the frontal sinus.

Agger nasi cells were reported to be present in more than 90% of the cases and frontal recess cells in more than 70% of the cases.^[5] This critical anatomical area is often diseased secondary to ethmoid infundibular disease which leads to hypoventilation resulting in epithelial changes like polypoidal degeneration or cystic dilatation of the mucosal glands.

Apart from the anatomic factor, occurrence of multiple mucocoeles, mucocoeles with acute infection, mucocoeles with significant extension outside the sinus walls, coexistent nasal polyposis, previous surgical injury in the frontal recess area also contribute for the recurrence of mucocoeles in the frontal recess area.^[6] High recurrence rate of mucocoeles are reported in multioperated patients with chronic sinusitis with a duration of a 2nd mucocoele after a mean interval of 4 years.^[7]

Inadequate marsupialisation with remnant cyst wall lining, retained medial superior and posterior wall of agger nasi cells, removing the normal lining mucosa, remnant frontal recess cells, retained uncinat process, middle turbinate lateralisation, osteogenesis and scarring are surgical reasons found contributing for the recurrence of disease in frontal recess area.^[8]

Radiological assessment of the frontal recess anatomy and its lesion is vital before surgery in the frontal recess area. Using CT scan, proper anatomical assessment of the three dimensional area of the frontal recess, the superior insertion of uncinat process, presence of frontal recess cells and its relation to agger nasi cell should be carried out using Kuhn classification.^[9] The radiological findings of a mucocoele in CT are homogenous isodense mass with clearly defined margin and patchy osteolysis around the mass. The extent of the lesion, its intracranial or orbital extension that can be determined by CT, aid in surgical planning. Identifying the plane of the lesion in frontal recess area is significant for the operating ENT surgeon. Requesting sagittal cuts for such lesions in and around the frontal recess area will be helpful to locate the plane of the lesion. Mucocoeles in MRI tend to be brighter than brain on T₁W images, and iso-hyperintense on T₂W images which is a pathognomonic MRI finding useful in differentiating from neoplasms and the neighbouring soft tissue structures involvement.^[10]

The surgical management of paranasal sinus mucocoeles require a customised approach ranging from endonasal endoscopic sinus surgery, external approaches like Lynch-Howarth, Caldwell-Luc, Osteoplastic flap approaches with or without sinus obliteration, Craniotomy or even a combined ENT-neurosurgical approach depending on the extension of the mucocoele.^[1] Transnasal endoscopic approach is often the first choice of treatment for frontoethmoidal mucocoeles since it is less invasive with less morbidity. Sinonasal endoscopic marsupialisation of frontal sinus mucocoeles was reported for the first time by Kennedy et al. in 1989.^[3] Marsupialisation improves the ventilation to the sinuses which helps to reverse epithelial metaplasia into normal respiratory epithelium.^[11]

With the progress in endoscopic techniques with intraoperative navigation, most lesions can be accessed with a minimally invasive, conservative and less aggressive approach than conventional external surgeries.^[12] However, external approaches and combined approaches are required for lateral frontal mucocoeles or mucocoeles with extensive intracranial or intraorbital extension.

Endoscopic surgical clearance of disease in the frontal recess area is technically challenging due to its anatomical variability, its location within the anterosuperior depths of the nasal cavity, and its close proximity to critical structures such as olfactory fossa, skull base, anterior ethmoidal artery and orbit.^[13] Multiple confounding factors for the recurrence of mucocoeles in the frontal recess area are identified as, adequate visualisation of the operative field with endoscopes optionally angled endoscopes, appropriate instrumentation, adopting the best approach to frontal recess, complete disease clearance, and a mucosal conservative technique.

Adequate marsupialisation to define the anterior boundary of frontal recess (anterior wall of agger nasi) by removing the rest of the agger nasi wall, removal of the entire cyst wall lining and leaving behind the normal mucosa without traumatising is essential to avoid future recurrences.

Using through-cutting instruments and powered microdebriders to preserve the mucosa in the sinuses will improve healing and avoid recurrences.^[14] The Messerklinger technique in endoscopic sinus surgery is designed to be minimally invasive to preserve the sinus mucosa and its physiological function. It is proved to be effective in dealing with even the most diseased frontal sinus.^[15]

Surgery on the frontal recess is preceded by an anterior ethmoidectomy followed by the clearance of disease in agger nasi cells to visualize the attachment of the middle turbinate medially, the lamina papyracea laterally and the anterior skull base with the anterior ethmoidal artery superiorly, as in most situations inflammatory changes in the frontal recess are secondary to ethmoidal infundibular disease.^[16] Stamberger's technique in frontal recess using angled endoscopes (30, 45 & 70 degree) and curved instruments is reported to be technically difficult by many authors. It is more difficult for type 3 cells with extension towards the skull base.^[9]

The anterior wall of agger nasi cells which is left intact during conventional angled endoscopic approach limits the exposure of the frontal recess. The agger nasi punch-out procedure is a safe technique, where removal of the anterior face of agger nasi cells improve the intraoperative exposure of the frontal recess even with a 0-degree endoscope. This surgical technique starts with complete uncinectomy including its superior one-third followed by anterior ethmoidectomy and maxillary antrostomy. A Hajek forceps is used to remove the bone and its overlying mucosa at the junction of the anterior attachment of the middle turbinate and lateral nasal wall in a vertical manner parallel to the middle turbinate so as to avoid destabilising the middle turbinate.

One or two bites are sufficient to remove the anterior face of agger nasi cells. An angled curette is used to curette the posterior wall of agger nasi cells from behind in a posterior to anterior direction away from the skull base. The bone fragments and soft tissue can be removed with an upbiting Blakesley forceps.^[13] This adequately enlarges the frontal recess area in the anteroposterior dimension and improves the ventilation of the sinus which helps to reverse the epithelial metaplasia into normal respiratory epithelium. Obstructive postoperative adhesions within the frontal recess may result due to this technique which is the cause of surgical failure in a few patients.^[13] Wormald (2002) claimed that the disadvantage of these approaches is the raw edge of the mucosa and bone that is left in the axilla of the middle turbinate, which can scar and pull the middle turbinate laterally, resulting in obstructive postoperative adhesions in the frontal recess.^[17]

The axillary flap technique designed by Wormald is designed to minimise the incidence of postoperative adhesions in the frontal recess by raising a posteriorly based mucosal flap and the raw area is wrapped at the end of the procedure.^[17] This technique starts with complete

uncinectomy followed by raising a posteriorly based full thickness flap from the lateral nasal wall that lies superior and anterior to the axilla of the middle turbinate.

It is important that the flap is elevated till behind the root of middle turbinate and reflected between the middle turbinate and septum exposing the anterior bony wall of the agger nasi cell. The anterior face of the agger nasi cell is punched out with Hajek forceps exposing the medial, posterior walls and the roof of agger nasi cell. An angled curette is used to remove the medial, roof and posterior wall of the agger nasi cell, which clears the frontal recess area. At the end of the procedure, the axillary flap is wrapped over the lateral side of the middle turbinate.^[8,17,18] This axillary flap approach was adopted for this patient, found to be easy, safe and gives excellent anterior access to the frontal recess area and aids in identifying frontal sinus ostium using 0 degree endoscope. This technique prevented scarring and postoperative adhesions in the frontal recess for 1-year follow-up period postoperatively.

CONCLUSION: In spite of multiple contributing factors for the recurrence of mucocoeles in the frontal recess, endoscopic axillary flap technique is found to be safe, easy with excellent anterior access to the frontal recess area preventing postoperative adhesions and scarring, thereby preventing recurrences.

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