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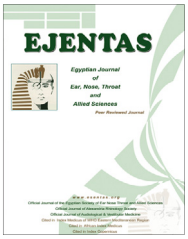

ORIGINAL ARTICLE

Frontocele: Experience from a resource challenged environment

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KEYWORDS

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Abstract *Background:* Frontocele commonly occurs as a result of obstruction in the outflow tract of the frontal sinus and this may be due to both congenital and acquired factors. Management involves the use of open, endoscopic or combined approaches with varying success and complication rates. *Objective:* This retrospective study highlights our experience with the management of frontocele in a resource challenged environment. *Methods:* A seventeen year retrospective analysis of all patients managed in our department was undertaken. Information was sourced from patient's case notes and operating theatre records. Data were analysed using Statistical Package for Social Sciences (SPSS) version 16 (SPSS Inc., Chicago, IL, USA) and Microsoft Excel 2007 (Microsoft, Redmond, WA, USA). *Results:* A total of 17 patients were managed within the years reviewed. Males accounted for 52.9% of the patients and ocular presentation was the commonest clinical presentation. Plain radiography alone was used in 76.5% of patients for assessment and bicoronal incision provided access to the frontal sinus in 88.2% of patients. Of the 17 patients managed, 1 (5.9%) patient died 24 h postoperative while 2 (11.8%) patients presented with recurrence 1 year and 3 years postoperatively respectively. *Conclusion:* The tendency for patients in our environment to present with extensive disease and to default in their postoperative follow-up appointment may favor a more radical approach in the management of frontal sinus mucocele.

1. Introduction

The frontal sinus is a part of the paranasal sinuses that is situated between the outer and inner tables of the frontal bone and consists mostly of two unequal parts separated by a

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septum that is rarely in the median plane. The important relations of the frontal sinus are the anterior cranial fossa and the orbit. The sinus is lined by respiratory mucus membrane and drains through one frontonasal duct on each side of the septum into the hiatus semilunaris of the middle meatus.[1]

Conditions affecting the frontal sinus may be congenital (such as bifid frontal sinus septum) or acquired such as trauma, infection, allergy and tumours, and this may result in partial or complete obstruction of the sinus drainage system. Obstruction of sinus drainage (or duct of the mucus glands within the epithelial lining of the frontal sinus) leads to mucus retention which over time results in gradual destruction of the bony walls of the sinus by the expanding mucus collection. This collection of slow-growing, benign expansile mucus filled mass which on histopathology consists of cyst-like structures lined with respiratory epithelium is referred to as frontal sinus mucocele or frontocele.[2] Frontoceles may become secondarily infected, forming a pyocele[3] and may be associated with signs and symptoms related to encroachment or expansion into adjacent anatomic structures and spaces such as the orbit, nose and anterior cranial fossa. Mechanisms postulated for mucocele expansion include pressure erosion, and active bone resorption/regeneration through inflammatory mediator's activity, particularly cytokines.[2,4]

Frontocele has been variously classified into types I–V (based on the extent of the expansile mass), and into medial, intermediate and lateral frontoceles (based on the position of the expansile mass).[5,6] The classification of frontocele has surgical implications with regard to extent and type of surgical intervention.

Occurrences of frontocele have been documented in both sexes with varying sex ratios and have commonly been reported in the middle age group.[2,7] Patient presenting complain depends on the anatomic structures encroached upon by the expanding mass. Diagnosis involves both clinical and radiological assessments. Treatment for frontocele is surgical and the approach may be external or endoscopic with varying reported success rates.[7,8]

This retrospective study highlights our experience in the management of seventeen patients in a resource challenged environment.

2. Patients and method

All patients who were managed for mucocele of the frontal sinus at the oral and maxillofacial surgery department of the Ahmadu Bello University Teaching Hospital, Shika-Zaria between September, 1997 and June, 2014 were retrospectively studied. Information was sourced from patient's case notes and operating theatre register. Information retrieved included age, sex, presenting complaints and duration, imaging technique used, surgical approach, duration of hospital stay and complications. We classified the clinical presentation of frontal sinus mucocele into frontal, nasal, ocular and intracranial. Data retrieved were analysed using Statistical Package for Social Sciences (SPSS) version 16 (SPSS Inc., Chicago, IL, USA) and Microsoft Excel 2007 (Microsoft, Redmond, WA, USA). Absolute numbers and simple percentages were used to describe categorical variables. Quantitative variables were described using measures of central tendency and measures of dispersion as appropriate. Test of statistically significant relationship was set at a *P*-value less than 0.05.

3. Results

A total of 17 patients were managed for mucocele of the frontal sinus within the period reviewed and consisted of 9 (52.9%) males and 8 (47.1%) females, giving a male to female ratio of 1.1:1. Patient's age ranged from 12 to 90 years with a mean age

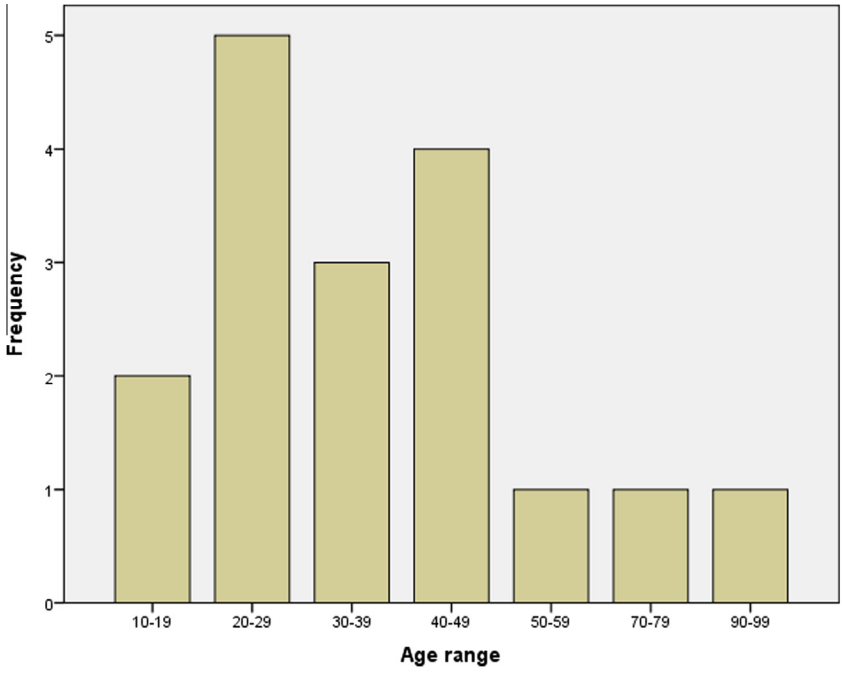

Figure 1 Age range distribution.

Table 1 Clinical presentation of frontocele based on site involved.

Clinical presentation	Frequency	Percentage
Ocular		
Proptosis with inferior-lateral displacement of the globe	14	23
Loss of vision	3	5
Exposure keratitis	4	7
Visual disturbance	11	18
Epiphora	4	7
Nasal		
Nasal blockage	2	3
Frontal		
Headache	5	8
Fluctuant frontal swelling	16	27
Frontocutaneous fistula	1	2
Intracranial	–	–
Total	60	100

of 37.4 ± 20.0 years. The highest incidence (29.4%) was recorded in the 20–29 year age group, followed by the 40–49 and 30–39 year age groups respectively (Fig. 1). Ocular presentation (60.0%) was the commonest clinical presentation with proptosis accounting for 38.9% of all ocular signs/symptoms (Table 1). One patient presented with bilateral proptosis of the globe. Of the 17 patients studied, 11 (64.7%) had documentation of site of involvement and of these, 7 (54.5%) patients presented with left facial symptoms, 4 (36.4%) had right facial symptoms while 1 (9.1%) patient presented with bilateral facial symptoms. The duration of presenting complain ranged from 12 months to 72 months with a mean of 25.6 months. There was no statistical significant relationship between duration of symptoms and clinical features ($P = 0.23$). Only 3 (17.6%) patients had documented known predisposing factors and these were trauma, craniofacial fibrous dysplasia and human immunodeficiency virus (HIV) associated immune defect respectively. The imaging modality used in assessing the frontal sinus and adjoining areas was plain radiography in 13 (76.5%) patients and computed tomography in 4 (23.5%) patients (Fig. 2).

The surgical incision used in accessing the frontal sinus via frontal craniectomy was bicoronal incision in 15 (88.2%) patients, extended hemi-coronal in 1 (5.9%) patient and extended temporal incision in 1 (5.9%) patient. All the patients had drainage of frontal sinus collections and removal of sinus lining with no obliteration. However, re-establishment of frontonasal drainage using a frontonasal tube was documented only in 5 (29.4%) patients. In addition, 2 (11.8%) patients had orbital evisceration by ophthalmologist while 1 (5.9%) patient had duroplasty by neurosurgeons for a defect in the posterior wall of the frontal sinus involving the duration.

The histological findings from specimen were documented only in 2 (11.8%) patients and these were chronic inflammation and fibrous dysplasia respectively. The duration of admission ranged from 12 days to 72 days with a mean of 23.3 ± 16.9 days. There was no statistical significant relationship between duration of symptoms and hospital stay ($P = 0.26$).

Of the 17 patients managed, 1 (5.9%) patient died 24 h postoperative while 2 (11.8%) patients presented with recurrence 1 year and 3 years postoperatively respectively. The mortality recorded was in the patient with HIV disease.

4. Discussion

Mucocele of the frontal sinus is associated with functional, aesthetic, social and psychological problems because the

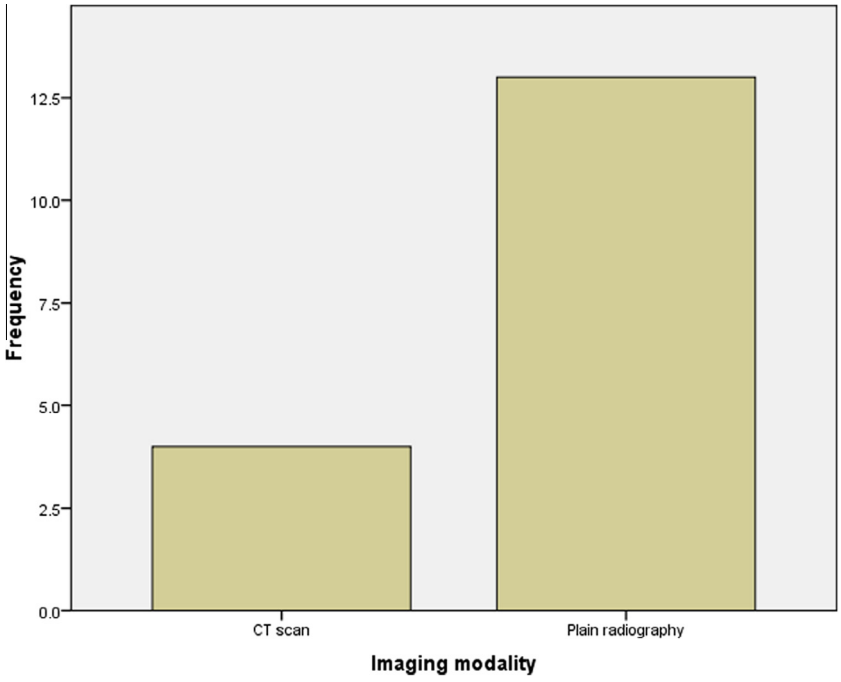

Figure 2 Imaging modality used in patient assessment.

anatomic region affected contain important structures and it is readily visible to the eye. A male to female ratio of 1.1:1 recorded in this study is less than the report from other studies.[7,9] However, a slightly higher female to male ratio has also been reported.[10] The higher male preponderance observed in most studies may be related to some anatomical differences between the male and female frontal sinuses. In one study, a statistically significant difference was found for mean total number of loculations in frontal sinus between males and females.[11] Similarly, males generally have slightly larger sinuses than females.[12,13] Trauma which has also been established in the aetiology of frontocele is known worldwide to occur more in males compared to females. These factors may account for the increased incidence seen in males.

The patient age in this study ranged from 12 to 90 years with most cases occurring in the 21 to 49 year age group, similar to other findings.[9] However, some previous reports documented a higher incidence in the 40–60 year age group.[10,14] Generally frontoceles can occur in any age group, although they are less frequently seen in children and the elderly. Occurrence of frontocele in the paediatric and elderly population is well documented in a few studies and case reports.[15–17] Of the seventeen patients we studied, there were two elderly and one paediatric patient.

Both congenital and acquired factors have been implicated in the aetiology of frontal sinus mucocele. These include trauma, chronic infection, tumours, allergy, aberrant sinus anatomy and systemic diseases such as cystic fibrosis,[7,18] however, some are idiopathic. Frontal mucocele may occur several years following exposure to any of these risk factors and this calls for a life time follow-up of patients.[19] Only three patients in our study had known predisposing factors and these were trauma, craniofacial fibrous dysplasia and human immunodeficiency virus infection. Frontal mucocele associated with fibrous dysplasia have been infrequently reported and may arise as a result of involvement of the frontal sinus recess by the dysplastic process.[20,21] HIV associated frontal mucocele is thought to arise from an immune reconstitution inflammatory syndrome following use of highly active antiretroviral therapy (HAART).[22] Similarly, HIV is associated with changes in mucociliary clearance and increased IgE mediated allergic events such as allergic rhinitis.[23,24]

The clinical presentation in mucocele of the frontal sinus depends on the anatomical structures encroached upon and can be classified into frontal, nasal, ocular and intracranial presentations. Ocular presentation includes proptosis (Fig. 3), visual disturbance, epiphora, altered papillary level and diplopia while nasal presentations include nasal blockage and discharge. Frontal headache, frontal swelling and frontocutaneous fistula are some of the frontal manifestations. In this retrospective analysis, ocular presentation had the highest frequency and this is similar to reports from other studies.[7,25] None of the patients in this study presented with intracranial manifestation. Epidural abscess, meningitis, subdural empyema, brain abscess, cerebrospinal fluid fistula and cranial nerve palsies are some of the intracranial manifestations.[26] However, some ocular manifestations such as visual disturbance may be due to intracranial extension. Similarly, the differential diagnosis of the various clinical manifestations should always be borne in mind. Some of the differential diagnoses of frontal mucocele are paranasal sinus carcinoma, aspergillus infection, chronic infection or inverting papilloma, dysthyroid eye disease, retrobulbar orbital tumour, inflammatory pseudo tumour and metastatic lesions.[26]

Figure 3 Right proptosis with inferior lateral displacement of the globe.

Both clinical and radiologic examinations are essential in the diagnosis of frontal mucocele since other sinus pathologies may present with similar features depending on the stage of the disease. Clinical examination will require interdisciplinary collaboration involving the ophthalmologist, maxillofacial surgeon/otorhinolaryngologist and the neurosurgeon. This is important for optimal surgical planning and for medico-legal reasons. Radiological examination involves the use of plain radiography and ultrasonography or advanced imaging modalities such as CT scan and magnetic resonance imaging (MRI). Findings on plain radiographs (occipitomental, posterior–anterior and lateral skull views) may include expansion and opacity of the sinus, attenuation of the normal thin mucoperiosteal white line, presence of dense reactive bone around involved sinus, loss of the normal scalloping of the sinus due to erosion of the septa and displacement of intersinus septum, erosion and sclerosis of the orbital wall(s) and obliteration of the nasal cavity. Calcifications may be present or absent.[27,28] Ultrasound scan may demonstrate the mucocele as a homogenous hypoechoic mass and is also useful in orbital assessment.[3] CT scan and MRI give detailed information on the hard and soft tissue (bone, dura, orbital soft tissues, etc) characteristics. CT scan unlike MRI provides more detailed information on the bone condition around the mucocele while MRI allows for a detailed evaluation of the soft tissue structures around the lesion. Generally, the appearance on CT and MRI depends on the degree of hydration of the mucocele. Inspissated mucocele appears hyperdense on noncontrast CT (Fig. 4) and hypointense signal on T1-weighted images, which becomes an area of signal void on T2-weighted images on MRI. Whereas a hyperintense signal on both T1 and T2 type images is consistent with a more hydrated secretion and these usually have a high protein content.[29] However, these features may be influenced by the presence of co-existent pathologies (Fig. 5). Only four of the patients in this study had CT scanning done, the remainders were evaluated using plain radiographs. The high use of plain radiographs is due to the following factors; non-availability of CT scan machine in our center prior to the year 2004, and inability of patients to afford the cost of CT scanning.

The treatment of frontocele is surgical and may be conservative or radical in approach. The aim of surgical management is to re-establish adequate drainage of the sinus and improve functional and aesthetic deformity. Conservative treatment

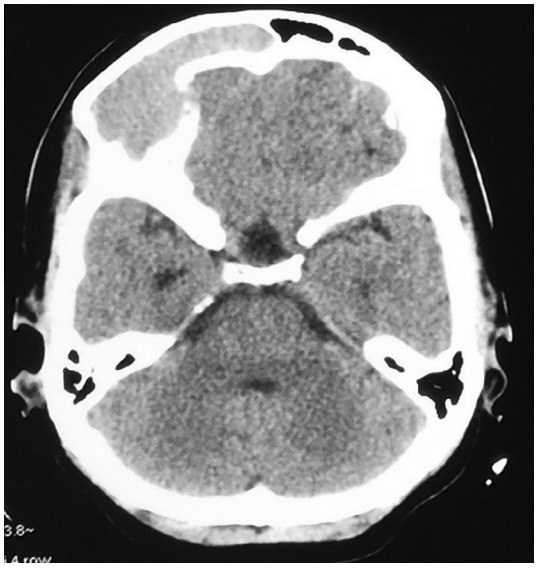

Figure 4 CT axial scan showing Hyperdense Frontal sinus in frontocele.

involves the use of functional endoscopic sinus surgery (FESS) for marsupialization of the lesion while the more radical approach involves an external or open approach. A combination of external and endoscopic approaches is also possible. The approach employed may be influenced by surgeon or patient's preference, surgeon's skill, available surgical tools, site and extent of the lesion among other factors. The external approach allows for complete exposure of the sinus (Fig. 6), provides complete obliteration of the sinus and prevents blind curettage of any exposed duramater.[26] On the other hand; FESS approach avoids leaving a scar and offers a safer and more definitive option than the open approach. However, in a meta-analysis of contemporary management of frontal sinus mucoceles, the external, endoscopic and combined approaches had similar recurrence rates.[30] All the patients in this retrospective study were managed using the external approach with an improvement in symptoms (Fig. 7). In most of our cases, on exposing the frontal bone, there usually was an area of bone dehiscence which was carefully widened and the cystic mass cleared. The external approach is generally favoured in our environment[16,17,28] and this may be due to the extent of lesion and lack of skilled manpower/instrumentation for FESS.

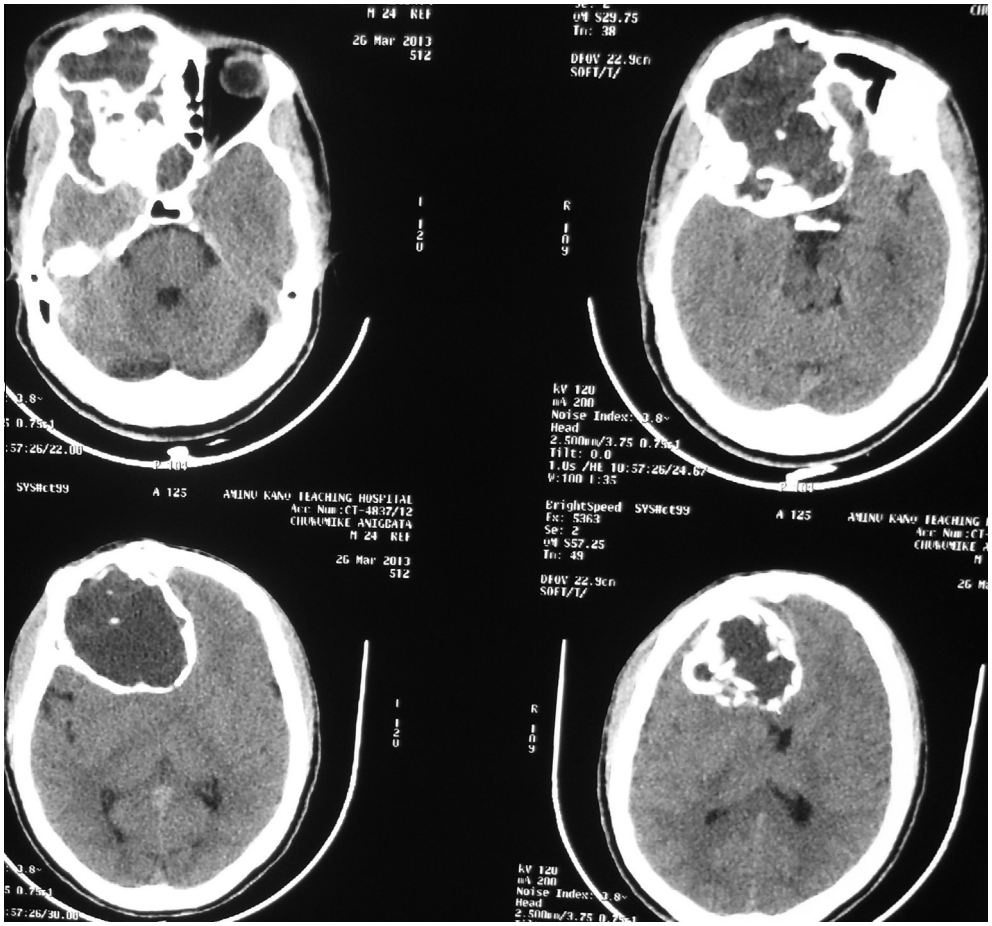

Figure 5 CT axial scan showing mixed densities in the right frontal sinus and anterior cranial fossa in frontocele secondary to craniofacial fibrous dysplasia.

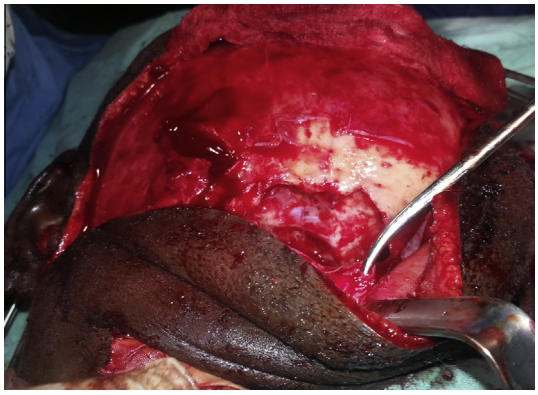

Figure 6 Exposure and clearance of the frontal sinus via bicoronal approach.

Figure 7 Improved proptosis post surgery.

Postoperative complications noted in this study were recurrence in two patients and mortality in one patient who was HIV positive. The recurrences were noted at one and three years postoperatively respectively. The cause of mortality is unknown since there was no autopsy performed because of the religious belief of some who were Muslims and others could not afford the cost.

Patient's response to follow up was poor and only four patients presented for follow-up. This may be due to the feeling of wellbeing or financial constraint (this affects their ability to transport themselves to the hospital). This has been a major problem in assessing treatment outcome in our environment. Hopefully, the availability of affordable mobile communication system in the near future in our environment will bring about a great improvement.

5. Conclusion

Although different approaches exist for the management of mucocele of the frontal sinus, surgeon's skill, availability of instrumentation, extent and site of disease and presence of co-existing pathologies are some of the factors that will determine the preferred approach in a particular environment. Despite the limited resources in our environment, the management of frontocele with available skills and facilities has been rewarding to both surgeons and patients. The tendency for patients in our environment to present with extensive disease and to default in their postoperative follow-up appointment may favour a more radical approach in the management of frontal sinus mucocele.

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Conflict of interest

None.

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