Case Report

Bilateral orbito-cerebral-extending frontal mucocele following nasosinus polyposis: A case report

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ABSTRACT

Introduction: and importance: Mucoceles are expansive pseudocystic formations, developed from the sinuses of the face, affecting mainly adults. Evolving at low noise, they are most often revealed by neurological or ophthalmological complications. We report a rare case of a bilateral frontal mucocele with orbito-cerebral extension following nasal sinus polyposis.

Case presentation: This was a 35-year-old patient with a history of Widal syndrome, who presented frontal headaches and left proptosis evolving for 4 months, in whom clinical examination revealed a left superomedial eyelid swelling, left proptosis and stage 2 nasosinus polyposis. Computed tomography and craniofacial magnetic resonance imaging were in favor of a bilateral frontal mucocele with left orbital and bilateral cerebral extensions. The patient was bilaterally operated by a combined approach including external Jacques eyebrow and endonasal Draf IIa procedure in addition to a radical total ethmoidectomy. The outcomes were favorable with regression of headaches and resolution of exophthalmos.

Clinical discussion: The frontal mucocele, although benign, has an aggressive potential in the absence of treatment either towards the endocranium or the orbit behind the orbital septum causing intra-orbital extension, or in front of it; causing the dominant upper palpebral form as in the case of our patient. The treatment is still based on surgical excision of the cyst with drainage of the causal sinus, which was carried out for our patient.

Conclusion: Despite its benign behavior, frontal mucocele may become serious by compression of neighboring organs which require an early and appropriate surgical management.

1. Introduction

Mucoceles are expansive cystic pseudotumors of the paranasal sinuses limited by a respiratory epithelium and filled with mucus, following the obstruction of the causal sinus drainage pathways [1]. Hence, their progression is very low and often asymptomatic. Therefore, this is associated with a diagnostic delay as they are only discovered at the stages of serious orbital or endocranial complications [2,3]. This rare tumor involves frequently the frontoethmoid complex [1] and it rarely extends to the endocranial anatomical components [4]. In this paper, we report a case of bilateral frontal mucocele with orbito-cerebral extension following nasosinus polyposis and we discuss this presentation based on a review of the literature. The current case is reported as recommended by SCARE guidelines [5].

2. Case presentation

Our patient was a 35-year-old male with a history of Widal syndrome who presented frontal headache, resistant to the usual analgesics associated with a left proptosis that progressed for 4 months. The ophthalmological examination found a left superomedial eyelid swelling, left proptosis and stage 2 nasosinus polyposis. Computed tomography and craniofacial magnetic resonance imaging were in favor of a bilateral frontal mucocele. The patient was bilaterally operated by a combined approach including external Jacques eyebrow and endonasal Draf IIa procedure in addition to a radical total ethmoidectomy. The outcomes were favorable with regression of headaches and resolution of exophthalmos.

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Several diagnoses were proposed including epidermoid cysts, cholesteatomas, meningiomas, chordomas, neurofibromas, and other tumors. MRI-based imaging was decisive in the diagnosis of the frontal mucocele. It can also assess the extension to the adjacent structures [13, 18] and also be associated as in the case reported in this paper. Thus, headaches and exophthalmos are characteristic of the frontal mucocele as in our case for the left frontal mucocele while that right was incidentally discovered on imaging. The development of these lesions is long and insidious varying between 2 months and 25 years [12]. In our case, this period was 4 months. Given its aggressive potential, a frontal mucocele can spread in the absence of treatment either towards the endocranial cavity by erosion of the posterior cortical wall of the frontal sinus which is the most serious but rare evolution and can led to meningoencephalic complications or rhinorrhea [4]. This entity can also extend to the orbital cavity by erosion of the anterior cortical wall which can be the origin of a lateralized exophthalmos. Moreover, these two locations can also be associated as in the case reported in this paper. Thus, headaches observed in our patient are due to the intracranial development of the mucocele exerting a mass effect on the frontal lobe. The mode of oculo-orbital extension of the frontal mucocele depends on the site of the anterior opening of the frontal sinus in relation to the orbital septum. If the opening is located in the frontal area, the frontal mucocele will extend further in the thickness of the upper eyelid like our case. On the other hand, if this opening occurs behind, the extension of the frontal mucocele will be inside the orbit. These two types of oculo-orbital mode of frontal mucocele extension can influence the treatment decisions and the expected outcomes. The craniofacial CT scan is now the firstline imaging procedure to diagnose the mucocele. It can also assess the associated bone lesions such as osteomas of the paranasal sinuses, craniofacial fibrous dysplasia and/or complications including abscess, intra-oral rupture, bone lysis, and brain involvement. According to the classification proposed by Thiggaranaj [17], our patient was categorized as type Va before erosion of the anterior and posterior walls of the sinus with minimal intracranial development. MRI, with better resolution of tissue contrast, is considered as the gold standard as it better specifies the extension to the adjacent structures [13, 18] and also allows the exclusion of differential diagnoses such as encephaloceles, epidermoid cysts, cholesteatomas, meningiomas, chordomas, neurofibromas, and polypoid tumors. The imaging workup is also of capital importance in the choice of the approach, and therefore for the multidisciplinary team involved in the treatment [19]. Surgery is the standard treatment of mucocele based on two approaches including external and endonasal routes or also via an endoscopic endonasal strategy. Several surgical techniques can be used for this purpose mainly those using gingivo-jugale route of Caldwell-Luc for maxillary mucoceles and the eyebrow route of Jacques for frontal or fronto-ethmoidal mucoceles as well as the bicoronal approach of Cairne Unterberger [16].

The treatment is based on the reintegration of the sinus housing the mucocele into the respiratory system (nasalization) or its exclusion by...
filling or cranialization. The principle of the external pathway is similar and is based on the large communication of the sinuses with the normal drainage system [20]. Endoscopic surgery by endonasal route is currently accepted as the method of choice in the management of mucoceles due to its low iatrogenicity and its excellent efficiency. This approach enables a respect of the healthy mucosa and a widening the natural drainage routes [21,22]. However, it should be indicated for lateral and extensive frontal localizations, skin fistulizations, and suspicion of associated malignant lesions and for the prevention of the risk of recurrence in the event of associated inflammatory ethmoidal pathology [23]. Our patient was operated bilaterally by a combined route using an endonasal approach by frontal sinusotomy through a Draf IIa in addition to a radical total ethmoidectomy given the failure of the well-conducted medical treatment of the nasosinus polyposis and also by using external approach. Long-term monitoring should be provided, as nasosinus mucoceles can recur several years after the initial surgery at a rate varying from 3 to 35% [24]. The 3-month follow-up of our patient is insufficient to judge the long-term evolution of the mucocele. However, the prognostic outcomes in these cases are most often good, with a resolution of the exophthalmos and a regression of headaches. The patient was satisfied with our management.

4. Conclusion

Frontal mucoceles are benign lesions. Their severity is due to their aggressive potential towards neighboring organs which may be associated with functional or even vital prognosis. Their management was improved with the advent of imaging and endonasal surgery which is considered as the gold standard. In our setting, external surgery seems desirable as these cases are diagnosed at advanced stages with endocranial expansion.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

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Author contribution

Dr Khalid Bouhafs: writing the manuscript, Review and Editing.
Professor Azeddine Lachkar: Visualisation, Conceptualisation and Investigation. Dr Tayeb Bouamama: provided the imaging data of the
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Research registration

N/a.

Guarantor

The Guarantor is the one or more people who accept full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

Declaration of competing interest

The authors have no conflict of interest to declare.

References

[1] H. Benkhatr, A. Gaultier, P. Halimi, P. Bonfils, Mucoceles sinusiennes et pneumo-sinus dilatans, Encycl Med Chir Orto Rhino Laryngol 11 (4) (2016) 20-481. B-10.
[2] J. Kouassi-N'djeundo, F. Buraïma, V. Ngattia, K.E. Badou, B.T.S. Vrob, M. Yoda, Ekrak, Les mucoceles sinusiennes: 2nde épidémio-clinique et thérapeutique de 25 cas colligés à Abidjan (Côte d'Ivoire), Mali medical 30 (3) (2015) 1-6.
[3] R. Zainine, I. Loukil, A. Dhaouadi, M. Ennaili, A. Mediouni, H. Chahed, et al., Complications ophtalmologiques des mucoceles rhino-sinusiennes, J. Fr. Ophthalmonl. (2014) 93-98.
[4] T. Obedobe, F. Olege, S. Segun-Busari, D. Nzech, Recurrent bilateral fronto-ethmoidal mucocele with intracranial extension: a case report, W. Afr. J. Med. 24 (3) (2005) 268-271.
[5] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, Scare Group, The SCARE 2020 guideline: updating consensus surgical CaseRepor (SCARE) guidelines, Int. J. Surg. 84 (2020) 226-230, https://doi.org/10.1016/j.ijjsu.2020.10.034.
[6] P.J. Conboy, N.S. Jones, The place of endoscopic sinus surgery in the treatment of paranasal sinus mucoceles, Clin. Otolaryngol (2003) 207-216.
[7] A.K. Mahapatra, S. Gaikwad, C. Sarrar, Giant mucoceles of the frontal sinus, J. Neurochir. 102 (1990) 114.
[8] R. Malhotra, P.J. Wormald, D. Selva, Bilateral dynamic proptosis due to fronto-ethmoidal mucocoele, Ophtalmol. (2014) 93.
[9] H. Manaka, K. Tokopo, K. Sakata, A. Ono, I. Yamamoto, Intracranial extension of mucocele complicating fronto-ethmoidal sinus osteoma: case report, Surg. Neurol. 50 (1998) 453-456.
[10] B. Verillard, J.P. Blanca, R. Kania, P. Herman, N. Le Clerc, J.P. Guichard, M. Classe, Mucocele formation after surgical treatment of interened papilloma of the frontal sinus drainage pathway, Am J Rhinol Allergy 30 (5) (2016) 181-184.
[11] H. Manaka, K. Topoko, K. Sakata, A. Ono, I. Yamoto, Intradural extension of mucocele fronto-ethmoidal sinus osteoma: case report, Surg. Neurol. 50 (1998) 453-456.
[12] I. Ndiaye, E.M. Diop, R. Diouf, N.D. Ndiaye, T. Ndomage, A. Tall, P.A. Ndiaye, Mucoceles du sinus frontal. A propos de 35 cas, Dakar Med. 39 (1994) 143-147.
[13] S. Peric, C. Sequet, J. Cabanes, A. Viosot, D. Kratinova, P. Derome, F. Chabolle, Mucocèles frontales à extension orbitaire ou cérébrale: stratégie thérapeutique, Anotolaryngol Chir Cervicofac 113 (1996) 384-391.
[14] A. Rivron, J. Bourdinière, Mucoceles et pneumo-sinus dilatans EMC, ORL (Oto-Rhino-Laryngol.) (Basel) 20465 A (10) (1990) 12.
[15] R.A. Benedict, M.K. Roth, V.N. Ghana, D.C. Brown, C.A. Geyer, Spontaneous drainage of an ethmoidal mucocele; a possible cause of pneumosinus dilatans, AJNR Am J Neuroradiol 12 (4) (1991) 729-731.
[16] G. Molteni, R. Spinelli, S. Pargatti, L. Colombo, P. Ronchi, Voluminous fronto-ethmoidal mucocele with epidual involvement. Surgical treatment by coronal approach, Acta Otohynolaryngol Ital 23 (2003) 185-190.
[17] Thiagarajan B. Fronto-ethmoidal mucocele within tracranial extension, an interesting case report and literature review. Internet version & ENT Scholar [Internet]. 24 mai 2012;Disponible sur: http://entscholar.wordpress.com/article/frontoethmoidal-mucocele-within-intracranial-extension-an-interesting-case-report/.
[18] M. Raynal, R. Peynegre, R. Beauru, A. Coste, Mucoceles sinusiennes et iatrogénie chirurgicale, An Otolaryngol Chir Cervi Fac 116 (1999) 85-91.
[19] A. Sama, L. McClellan, J. Constable, Frontal sinus mucoceles: new algorithm for surgical management, Rhinology 52 (2014) 267-275.
[20] A. Pompili, R. Mastrosteфано, F. Cardì, F. Cattani, F. Cianfriglia, P.F. Nardis, et al., Mucoceles of neurosurgical interest: clinical considerations on five cases, Acta Neurochir. 102 (1990) 114-121.
[21] D. Kennedy, Functional endoscopic sinus surgery, Arch. Otolaryngol. 111 (1985) 643-649.
[22] H. Stammberger, Endoscopic endonasal surgery—concepts in treatment of recurring rhinosinusitis. Part II. Surgical technique, Otolaryngol. Head Neck Surg. 94 (1986) 147-156.
[23] Salf E. TrannoyPh, J.L. Poncet, P. Buffe, Y.F. Ludennec, Mucoceles sinusiennes. Modalités thérapeutiques (à propos de 6 cas), Cahiers d’O.RL 28 (1993) 172-177.
[24] BradleyNJP, N.S. Jones, Paranasal sinus mucoceles: modern management, AJR 9 (1995) 251-256.