OPEN ACCESS

Saudi Journal of Medical and Pharmaceutical Sciences

Abbreviated Key Title: Saudi J Med Pharm Sci
ISSN 2413-4929 (Print) |ISSN 2413-4910 (Online)
Scholars Middle East Publishers, Dubai, United Arab Emirates
Journal homepage: https://saudijournals.com/sjmps

Original Research Article

Naso-Sinusian Mucoceles: Retrospective Study of 24 Cases

Mohamed Ali Gliti[1,3*], Bencheikh Razika[2,3], Rahim Hanaa[1,3], Benbouzid Mohamed Anas[2,3], Leila Essakalli Houssyni[2,3]

[1]Resident Physician in Otorhinolaryngology, Department of Otorhinolaryngology, Head and Neck Surgery, Ibn Sina University Hospital, Rabat, Morocco
[2]Professor of Otorhinolaryngology, Department of Otorhinolaryngology, Head and Neck Surgery, Ibn Sina University Hospital, Rabat, Morocco
[3]Faculty of Medicine and Pharmacy of Rabat, Mohammed V University, Rabat, Morocco

DOI: 10.36348/sjmps.2020.v06i11.003 | **Received:** 26.07.2020 | **Accepted:** 03.08.2020 | **Published:** 06.11.2020

***Corresponding author:** Mohamed Ali Gliti

Abstract

Objective: Highlight the different clinical and surgical features of this pathology. Bring the experience of the service and the chirurgical specificities encountered during the care of these patients. ***Materiels and méthods:*** Retrospective study of 24 cases of paranasal sinus mucoceles collected and operated in our department since January 2017 to December 2019. ***Results:*** The average age was 41 years. We noted 22 cases of ethmoid-frontal localization, 1 case of maxillary localization and 1 case of sphenoidal localization. The most common symptoms were swelling of the inner corner of the eye and exophthalmia. Complicated superinfection of orbital cellulitis was revealing in 3 cases. Facial computed tomography confirmed the diagnosis in the majority of cases. Magnetic resonance imaging (MRI) was requested in 3 patients. The surgical treatment consisted of a wide marsupialization by the endo-nasal route in 24 cases and by combined approach in 1 case. A recurrence was observed in two patients, after an average delay of 12 months. ***Conclusion:*** Mucocele is a relatively rare benign pathology, its symptomatology depends on its location. CT and MRI of the facial sinuses represent the key exams for diagnostic assertion. Treatment is surgical. Endonasal endoscopic marsupialization is currently the treatment of choice.

Keywords: Paranasal sinus mucocele, swelling of the face, exophthalmia, marsupialization.

INTRODUCTON

The therapeutic approach to nasosinus mucoceles constitutes a therapeutic difficulty and challenge, and a certain number of approaches have been reported over the years.

Generally, external approaches give broad exposure, but can be complicated by scarring and nerve and skin-mucosal damage, while endoscopic approaches are limited because lateral and suprorbital extensions of the sinus may be inaccessible.

MATERIELS AND METHODS:

This is a retrospective study of 24 cases of nasal sinus mucocele collected and operated on in the otolaryngology and head and neck surgery department from January 1, 2017 to December 31, 2019. The data collected included demographic data of patients, surgical details, pathological findings and complications. All procedures were performed under general anesthesia. All patients underwent preoperative and postoperative computed tomography of the orbit and para nasal sinuses.

RESULTS

The number of patients received in our service during these two years was 24 patients, the average age is 49 years ranging from 21 to 72 years, and the majority were male with a sex ration of 22H / 2F.

The history of trauma with point of craniofacial impact was found in all our patients.The average time between trauma and the onset of first symptoms was 3 years ranging from 1 year to 15 years. The most frequent mechanism is road accidents.

The most common symptom is swelling of the inner corner of the eye and exophthalmia in 98% of cases, followed by diplopia in 90% of cases, tearing in 56% and headache in 48%. The average consultation time between the appearance of the first symptoms and the first consultation is 4 years ranging from 1 year to 10 years.

The complicated secondary infection of orbital and pre-septal cellulitis was revealing in 3 cases, which required cooling antibiotics for them, based on rcephine 1 at a dose of 2g IVD per day for 10 days and metronidazole at a dose 500mg IVL every 8 hours for 10 days, the surgical intervention was scheduled 5 weeks later.

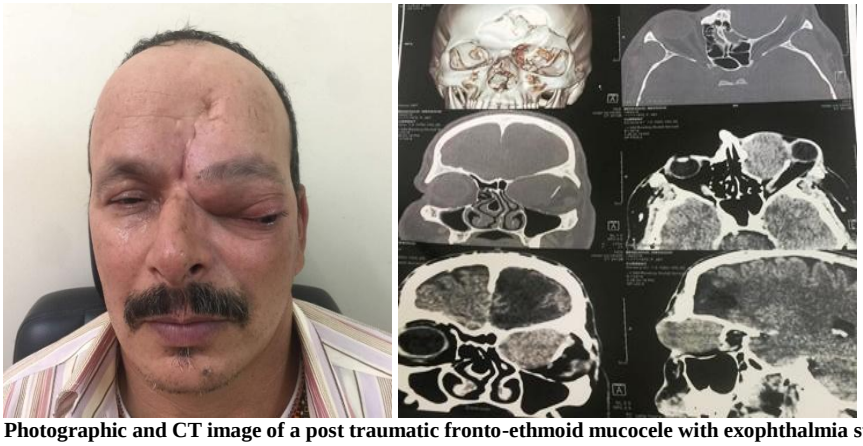

Fig-1: Photographic and CT image of a post traumatic fronto-ethmoid mucocele with exophthalmia stage 3

CT scan was performed in 100% of patients allowing confirmation of the diagnosis, as well as a well-established lesion assessment, while magnetic resonance imaging was performed in three cases where endocranial extension was doubtful in one patient, and to better visualize the relationship with the eyeball in two cases.

We noted 22 cases of ethmoid-frontal localization, 1 case of maxillary localization and 1 case of sphenoidal localization. The majority of patients presented with unilateral locations: 56% left and 44% right.

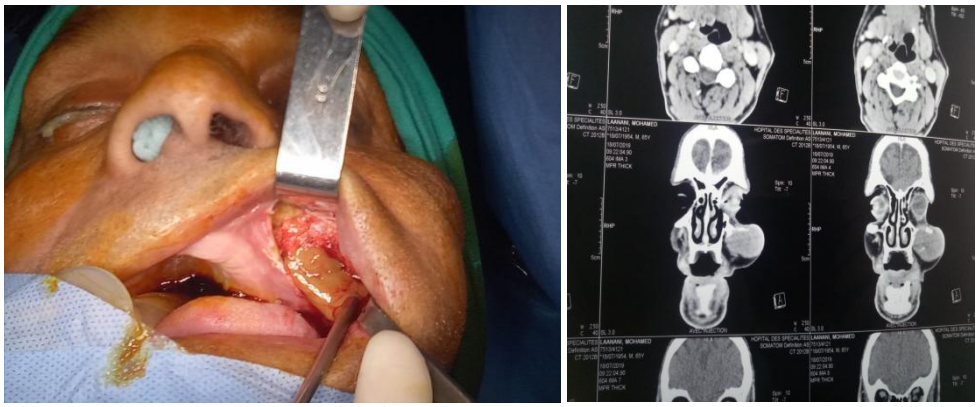

Fig-2: CT and intraoperative images of a left maxillary mucocele treated by the combined route (Caldwel-Luc)

The surgical treatment consisted of a wide marsupialization by the endo-nasal route in 24 cases and by combined approach in 1 case. No patient had immediate post-operative complications. A recurrence was observed in two patients, after an average delay of 13 months. Table-1 summarizes the clinical data of patients from our series

Fig-3: Photo of a patient showing a recurrence of the fronto ethmoid mucocele 6 months after the first intervention

Table-1: Clinical data of patients from our series

	Age	Sex	Medical history	Consultation dela (month)	Localisation	Clinical signs	Type of surgery	Duration of follow-up (month)	Recurrence
1	40	Male	Craniofacial trauma	14	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	6	-
2	39	Male	Craniofacial trauma	22	ethmoid-frontal	Exophtalmia	endo-nasal	4	-
3	46	Male	Craniofacial trauma	16	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	9	-
4	21	Male	Craniofacial trauma	34	ethmoid-frontal	Exophtalmia+diplopie+ tearing+headach+surinfection	endo-nasal	10	After 2 months
5	54	Male	Craniofacial trauma	42	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	12	-
6	48	Male	Craniofacial trauma	21	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	19	-
7	63	Male	Craniofacial traumahigh blood pressure+ Diabetes	14	Maxillary	Exophtalmia+diplopie+ tearing+headach	combined approach	24	-
8	42	Male	Craniofacial trauma	12	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	12	-
9	45	Male	Craniofacial trauma	36	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	5	-
10	40	Male	Craniofacial trauma	24	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	6	-
11	51	Female	Craniofacial trauma+ high blood pressure	12	Sphenoidal	Exophtalmia+diplopie+tearing +headach	endo-nasal	8	-
12	47	Male	Craniofacial trauma	48	ethmoid-frontal	Exophtalmia+diplopie+tearing +surinfection	endo-nasal	24	After 4 months
13	62	Male	Craniofacial trauma	72	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	12	-
14	42	Male	Craniofacial trauma	48	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	8	-
15	60	Male	Craniofacial trauma+ Diabetes	24	ethmoid-frontal	Exophtalmia+diplopie+tearing	endo-nasal	12	-
16	38	Male	Craniofacial trauma	72	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	36	-
17	72	Male	Craniofacial trauma+ high blood pressure +Diabetes	180	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	11	-
18	43	Male	Craniofacial trauma	24	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	12	-
19	56	Male	Craniofacial trauma	48	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	7	-
20	64	Male	Craniofacial trauma	60	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	18	-
21	45	Female	Craniofacial trauma	24	ethmoid-frontal	Exophtalmia+diplopie	endo-nasal	12	-
22	52	Male	Craniofacial trauma	60	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	4	-
23	47	Male	Craniofacial trauma	24	ethmoid-frontal	Exophtalmia+ surinfection	endo-nasal	36	-
24	58	Male	Craniofacial trauma	36	ethmoid-frontal	Exophtalmia+diplopie+tearing +headach	endo-nasal	10	-

DISCUSSION

The average age at diagnosis and the male predominance in our series are comparable to those reported in the literature [8, 13, 14, 18].

In our series, the most common location is that of the ethmoid or frontal sinuses, consistent with comparable series in which the frequency varies from 65% to 80% [8, 13, 18, 2].

The maxillary localization of the mucocels in our series (4.1%) is less frequent compared to that recorded in the non-oriental series, in which it varies between 5.5% and 25% [8, 13, 18].

As well as localization in the sphenoid sinus (4.1% in our series), considered to be the least frequent in the literature, in which it varies from 6% to 11% [8, 13, 18].

In the series of Sergio O *et al*., [19], a single maxillary mucocele was treated in the time preceding the introduction of CT in their center. They believe that many maxillary mucoceles remained underdiagnosed, by the lower specificity and clinical exuberance of the early stages compared to the frontal or ethmoid mucoceles. In 6% of cases, there was a multiple sinus assignment, while in the previous series, the available frequency was between 3% and 16% [8, 13, 18].

All the patients in our series presented a craniofacial trauma explaining the occurrence of mucoceles. In the series of Sergio O *et al*., [19], Twenty-nine per cent of the patients presented no factor that could justify the appearance of mucoceles, a frequency higher than that reported in certain series, varying between 7% and 18% [8, 13, 14]. The highest frequency of primary mucoceles was recorded in the series of Lund *et al*., [17], with 52%, although this series does not include the maxillary mucoceles, records only 48 cases and its results are not reproduced in other series.

In the series of Sergio O *et al*., [19] Thirty-five percent of the patients in their series had a history of nasal sinus surgery, with a predominance of the external route over the endoscopic route, as is the case in most [13, 19]. In the most representative series, the frequency of surgical history is between 35% and 66% [8, 13], with an average latency between surgery and the diagnosis of mucocele of between 2, 8 and 24 years [13, 16].

In our series, the latency between the trauma and the diagnosis of mucocele is 4 years. In the series by Sergio O *et al.*, [19], the latency between the surgical history and the diagnosis of mucocele was 15 years for the group with a history of external surgery and 5 years for the endoscopic surgery group. The increased latency for mucocele development after open approaches is confirmed by other studies [13, 16], although we must bear in mind that these data are very falcified, given the late development of endoscopic surgical techniques. DeFreitas *et al*., [19] have highlighted the long latency period existing for the development of the maxillary mucoceles after Caldwel-Luc type surgery, with an average of 23 years.

88% of patients in our series had a history of frontal or nasal bone fracture. Only in the series of Sergio O *et al*., [19], only 14% of patients who had the same type of traumatic history, with an average latency of 25 years, were more frequent in the frontal and ethmoid mucoceles.

The rate reflected in the series, which includes mucoceles in any localization of the sinuses, varies between 1.6% and 14% [8, 13, 14] although in the series of Naudo *et al*., [19] with a small number of cases, history of trauma appears in 18% of patients.

In the series where the mucoceles are predominantly or exclusively frontal, the history of traumatic bone fracture is between 13% and 30% [15, 27]. However, there are several series with a high frequency of frontal affection and a low frequency of trauma history [13, 17]. In our series and that of others [8, 13, 14] link traumatic events of higher frequency to the frontal and ethmoid mucoceles.

No patient in our series had nasal polyposis, while in the series of Sergio O *et al*., [19] 31% of patients had polyposis, a frequency higher than that of the other series (6% -28%) [13, 14, 19]. In their series, polyposis was more frequent in the sphenoid mucoceles than in the maxillary, ethmoidal or frontal mucoceles.

In our series no diagnosis is accidental, in the series of Sergio O *et al*., [19] up to 6% of patients were diagnosed accidentally by computed tomography or MRI which was performed for another rhinological symptomatology, and accidental diagnosis was more frequent in the case of maxillary mucoceles, which remain asymptomatic for long periods.

Until the availability of CT scan, few maxillary and sphenoid mucoceles were diagnosed and many mucoceles from other locations were subjected to surgical investigations for suspected neoplastic disease. In the article by Marks *et al*., 11 to 21% of the maxillary mucoceles were diagnosed by chance. Series have been published in which, even in mucoceles affecting the orbital or cranial base, up to 8% have been diagnosed accidentally [11, 15]. We postulate that one of the causes which justifies the increase in the incidence of mucoceles in recent years is the development of better diagnostic techniques.

Loss of visual acuity occurs due to intra-orbital compression caused by mucocele, either acute or similar to post-septal or chronic cellulite. In the series by Sergio O *et al.*, [19] a patient with a sphenoid mucocele suffered from compressive optic neuritis. In the literature, the resolution rate for loss of visual acuity is very high (10/11) after surgical treatment [9, 11, 15]. Blindness and pupillary non-response are considered to be factors of poor prognosis for visual recovery and surgical treatment, as soon as possible, is favorable.

The total incidence of recurrence was 4.1% in our series in a patient operated by the endoscopic route, compared to 10% of patients in the series of Sergio O *et al.*, [19]; there were recurrences in 17% of patients operated on externally or in combination and in 5% of

patients treated by endoscopy. There are many series of cases, which reproduce a lower incidence of recurrence in mucoceles treated endoscopically than in those treated by external or combined approaches.

The largest number of mucoceles treated exclusively endoscopically corresponds to the Har-El *et al.*, [18] study with 103 patients and an average follow-up of 4.6 years, in which only one recurrence was noted. This low incidence of recurrence is confirmed in other series [14, 7, 15].

In addition, in the series by Khong *et al.*, [19], which includes orbital mucoceles treated by endoscopy, a recurrence rate of 8.3% is recorded. There are several series with a small number of patients which include the maxillary mucoceles treated endoscopically without recurrences [6, 16, 7] and in the only pediatric series [7], with patients and the ethmoid and sphenoid mucoceles treated endoscopically, no recurrence is identified either [19].

Table-3 details the characteristics of the most representative series of mucoceles treated by endoscopy. Thus, in most of the series, endoscopic marsupialization does not lead to a higher incidence of recurrence.

Table-2: Incidence of recurrences in the most notable series which include mucoceles treated endoscopically

	Patients ESS, No.	Location	Follow-up, average, mo	Patients with recurrences, %
Conboy et al 2003[26]	44	All the sinuses	74[a]	9
Serrano et al 2004[13]	33	All the sinuses	47.4	0
Rombaux et al 2000[23]	132	All the sinuses	44[a]	2.2
Har-El et al 2001[18]	103	All the sinuses	55.2[b]	1
Khong et al 2004[14]	28	Maxillary, ethmoidal, and frontal	18	0
Moriyama et al 1992[20]	47	Ethmoidal and sphenoidal	12-120[c]	0
Sautter et al 2008[31]	57	Erosion of the orbit or the base of the skull	15[b]	0
Khong et al 2004[27]	15	Erosion of the orbit	16	20
Saito et al 2000[16]	21	Postoperative maxillary	12-60[c]	0
Busaba et al 1999[6]	13	Maxillary	10-66[c]	0
Caylakli et al 2006[22]	14	Maxillary	17.8	0
Benninger et al 1995[24]	15	Ethmoidal and sphenoidal	20	13
Hartley et al 1999[32]	7	Paediatric ethmoidal and sphenoidal	>12	0
Lund et al 1998[17]	20	Frontal, frontoethmoidal, and sphenoidal	34	0

[a]General follow-up, of patients treated both by endoscopy and by open surgery.
[b]Median.
[c]Only the interval of follow-up is available.

It is cited in the literature that loss of soft tissue bone support in frontoethmoidectomies 8], compartmentalization of the sinus mucosa, scarring of facial tissue in Caldwell-Luc [6] antrostomies and remnants of the frontal mucosa in obliterating frontal osteoplasties are the main postoperative risk factors that facilitate the development of secondary mucoceles. Recurrences were also more common in patients with polyposis, given the perpetuation of this condition involved in incorrect sinus ventilation.

In our series, no immediate postoperative complication was noted either by endoscopy or by the combined route. In the series by Sergio O *et al.*, [19], there were no major complications in the group treated with endoscopic surgery. In the series by Serrano *et al.*, [13] and Schaefer *et al.*, [18], there were 1% and 2.8% respectively of major complications. In 4% of endoscopic surgical procedures, there were minor complications, which, in different series, varied between 3.5% and 6% [13, 14, 19].

In the group treated by open surgery, major complications occurred in 10% of cases, all in the frontal or ethmoidal passages. Up to 43% of cases had minor complications, a higher number than that recorded in the Serrano *et al.*, [13] series (22%).

In our series, no late complications have been noted so far. In the series of Sergio O *et al.*, [19], the occurrence of sequelae was more frequent in the endoscopically treated group than in that treated by open or combined surgery, although most were clinically silent synechiae and represented a lower morbidity than follow-up in open approaches.

In the Serrano *et al.*, [13] series, there were no sequelae in the endoscopic approach, while in the endoscopic series of Khong *et al.*, [15], they were present in 10% (7% were synechial) and in the series de Schaefer *et al.*, out of 33 endoscopic procedures on the frontal sinus, synechiae occurred in 8.3% of cases. Although frontal paraesthesia is the norm in osteoplastic approaches to the frontal sinus, it usually disappears in 3 months [18].

CONCLUSION

The endoscopic therapeutic approach to mucoceles is a safer and more effective technique, with fewer recurrences than the external approach, in addition to being better tolerated and involving less cost due to postoperative complications.

The most common sequelae are nasal synechiae, often with no clinical impact. The sequelae of external approaches are more serious and also involve external scarring.

The lateral localisation of frontal sinus mucocel does not allow it access from anatomic point of view.

REFERENCES

1. Kariya, S., Okano, M., Hattori, H., Sugata, Y., Matsumoto, R., Fukushima, K., ... & Nishizaki, K. (2007). Expression of IL-12 and T helper cell 1 cytokines in the fluid of paranasal sinus mucoceles. *American journal of otolaryngology*, *28*(2), 83-86.
2. Patrocinio, L. G., Damasceno, P. G., & Patrocinio, J. A. (2008). Maxillary mucocele in a 4-month infant. *Revista Brasileira de Otorrinolaringologia*, *74*(3), 479-479.
3. Chobillon, M. A., & Jankowski, R. (2004). Relationship between mucoceles, nasal polyposis and nasalisation. *Rhinology*, *43*, 219-24.
4. Bockmühl, U., Kratzsch, B., Benda, K., & Draf, W. (2006). Surgery for paranasal sinus mucocoeles: efficacy of endonasal micro-endoscopic management and long-term results of 185 patients. *Rhinology*, *44*(1), 62-67.
5. Atasoy, C., Ustüner, E., Erden, I., & Akyar, S. (2001). Frontal sinus mucoceles: a rare complication of craneofacial fibrous dysplasia. *Clin Imaging*. 25:388–391.
6. Di Cicco, M., Costantini, D., Padoan, R., & Colombo, C. (2005). Paranasal mucoceles in children with cystic fibrosis. *International journal of pediatric otorhinolaryngology*, *69*(10), 1407-1413.
7. Serrano, E., Klossek, J. M., Percodani, J., Yardeni, E., & Dufour, X. (2004). Surgical management of paranasal sinus mucoceles: a long-term study of 60 cases. *Otolaryngology—Head and Neck Surgery*, *131*(1), 133-140.
8. Khong, J. J., Malhotra, R., Selva, D., & Wormald, P. J. (2004). Efficacy of endoscopic sinus surgery for paranasal sinus mucocele including modified endoscopic Lothrop procedure for frontal sinus mucocele. *The Journal of Laryngology & Otology*, *118*(5), 352-356.
9. Constantinidis, J., Steinhart, H., Schwerdtfeger, K., Zenk, J., & Iro, H. (2001). Therapy of invasive mucoceles of the frontal sinus. *Rhinology*, *39*(1), 33-38.
10. Har- El, G. (2001). Endoscopic management of 108 sinus mucoceles. *The laryngoscope*, *111*(12), 2131-2134.
11. Caylakli, F., Yavuz, H., Cagici, A. C., & Ozluoglu, L. N. (2006). Endoscopic sinus surgery for maxillary sinus mucoceles. *Head & Face Medicine*, *2*(1), 29.
12. Rombaux, P., Bertrand, B., Eloy, P., Collet, S., Daele, J., Bachert, C., & Claes, J. (2000). Endoscopic endonasal surgery for paranasal sinus mucoceles. *Acta oto-rhino-laryngologica belgica*, *54*(2), 115-122.
13. Chiu, A. G., & Vaughan, W. C. (2004). Management of the lateral frontal sinus lesion and the supraorbital cell mucocele. *American journal of rhinology*, *18*(2), 83-86.
14. Conboy, P. J., & Jones, N. S. (2003). The place of endoscopic sinus surgery in the treatment of paranasal sinus mucocoeles. *Clinical Otolaryngology & Allied Sciences*, *28*(3), 207-210.
15. Khong, J. J., Malhotra, R., Wormald, P. J., & Selva, D. (2004). Endoscopic sinus surgery for paranasal sinus mucocoele with orbital involvement. *Eye*, *18*(9), 877-881.
16. Herndon, M., McMains, K. C., & Kountakis, S. E. (2007). Presentation and management of extensive fronto-orbital-ethmoid mucoceles. *American journal of otolaryngology*, *28*(3), 145-147.
17. Sautter, N. B., Citardi, M. J., Perry, J., & Batra, P. S. (2008). Paranasal sinus mucoceles with skull-base and/or orbital erosion: Is the endoscopic approach sufficient?. *Otolaryngology—Head and Neck Surgery*, *139*(4), 570-574.
18. Llames, A. L., Pendás, J. L., Fente, V. S., Martínez, G. B., & Nieto, C. S. (2003). Osteoplastia frontal: nuestra experiencia. *Acta Otorrinolaringológica Española*, *54*(6), 429-434.
19. Obeso, S., Llorente, J. L., Rodrigo, J. P., Sánchez, R., Mancebo, G., & Suárez, C. (2009). Paranasal sinuses mucoceles. Our experience in 72 patients. *Acta Otorrinolaringologica (English Edition)*, *60*(5), 332-339.