Paranasal Sinus Mucocele

Clinical, imagistic and biochemical aspects

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Paranasal mucoceles are a type of cysts that evolve slowly and are asymptomatic; this poses a difficulty in diagnosing the patient because the symptoms can go unnoticed. The mucocele evolves unpredictably. On the one hand, it can become infected turning into pyoceles and on the other hand, it can invade important regions such as the orbital, cranial or genian regions, creating facial asymmetry. This is a retrospective case study of 37 patients diagnosed with sinus mucoceles, followed up by clinical examination and paraclinical tests such as CT and MRI scans. The biochemical components of the liquid from within the mucocele were analyzed and the following criteria were recorded: NaCl-, Cl-, Na+ and cholesterine as well as cellular components such as mastocytes, macrophages, hematocytes and leucocytes. In all cases, the treatment option was surgery with favorable post-operative and follow-up evaluation. The mucoceles that appeared post-operatively (maxillary and ethnoid sinuses) evolved more rapidly than the mucoceles that were induced byan external injury. Longer follow-up of operated patients permitted a more timely diagnosis of recurrences.

Keywords: mucocele, diagnosis, imagistic and biochemical aspects

The mucocele has a cystic structure made from a membrane that develops from the sinus mucosa and contains viscous mucus. Factors that can cause mucocele formation are inflammatory, tumoral and traumatic.

These cystic tumors were first described in 1900 [1] but received the name of *mucocele* in 1896 by Rollet [2]. The mucocele is more frequently located in the frontal sinuses, followed by the fronto-ethmoidal region, then by the maxillary and sphenoid sinuses [3-5].

The mucocele forms due to progressive obstruction of the mucocilliary drainage due to epithelial dislocation that can happen after surgery or post-fraumatically. A primary mucocele develops without any prior injury of surgical interventions and a secondary mucocele develops after external injury or post-operatively [3].

The mucocele is surrounded by a bony structure and because the mucocele accumulates mucus and obstructs the sinus ostium, the pressure exerted along with the biochemical changes remodels and erodes the bony structures around the mucocele.

Thus, increased pressure leads to bone erosion but the chronic inflammation of the sinus mucosa which blocks the sinus' ostium triggers the production of cytokines, collagenase and prostaglandins which in turn stimulate bone resorption[6-8].

The mucocele grows progressively and remodels the surrounding bony structure. Its presence may go unnoticed for long periods of time and becomes noticeable during periods of acute pain or infection. The next phase is the exteriorization phase. In this phase the region from which the mucocele developed appears deformed. Occasionally the debut of a mucocele is signaled by an oculo-orbital complication. The nose and paranasal sinuses are potential infected cavities [9] and even though the mucus within the mucocele is usually aseptic, it can become infected and turn into a pyocele.

Experimental part

Material and methods

This is a retrospective study on 37 patients with sinus mucoceles from the University Hospital ENT Clinic in Galati. Out of the 37 patients studied, 21 patients were male (56.76%) and 16 patients were female (4.24%). Patient age ranged from 21 to 78 years. Clinical, imagistic, chemical and mucus cellularity were analyzed as well as treatment outcome.

Clinically, 19 patients had frontal sinus mucoceles, 13 patients had ethmoidal sinus mucoceles and three other patients had maxillary sinus mucoceles. The last two cases could not be classified due to the size of the mucocele.

The patients' history revealed that 12 suffered an injury (surgical intervention or external trauma).

In order to analyze the chemical composition of the mucus within the mucocele, 10 out of the 37 mucoceles were punctured and the afferent mucus analyzed (NaCl-, Cl-, Na+ and cholesterine) as well as other cellular components.

Results and discussions

Clinically, the patients presented with:

maxillofacial pressure present in 30 cases;
headache in 24 cases;

- •unilateral respiratory trouble persistent in 12 cases; ·rhinorrhea in 6 cases;
- pain in the internal angle of the eye in 4 cases;

deformation in the region corresponding to the mucocele in 15 cases.

There were patients that presented with two or more symptoms enumerated above.

Out of the patients studied, 22 patients had chronic rhinosinusitis without polyps (CRS) and 7 patients had chronic rhinosinusitis with nasal polyps (CRSwNP).

The concentration of the different elements studied from the mucus is shown in table 1:

All authors have equally contributed to the article

	Minimum concentration	Maximum concentration	Average
	mg%	mg%	mg%
NaCl-	465	490	480
C1-	288	293	290
Na+	302	310	306
Cholesterine	82		

Table 1

NaCl- concentration between 465-490 mg%, with an average of 480 mg%.

Cl- concentration between 288-293 mg%, with an average of 290 mg%.

Na+ concentration between 302-310 mg%, with an average of 306 mg%.

Cholesterine concentration between 46-88 mg%, with an average of 82 mg%.

Cellular elements found were mastocytes, macrophages, hematocytes and leucocytes.

The treatment option in all cases was surgery by external approach with the extirpation of the mucocele membrane as well as maintaining drainage and aeration of the sinus.

Maintaining drainage of the sinus ostium by excising the denatured bone (osteitis) permits drainage of the sinus cavity which maintains aeration and prevents possible relapse of mucocele formation.

The mucocele is a round or oval shaped structure located in the paranasal sinuses and can extend locally to other regions. The mucoceles wall is made of a fibrous membrane, gray-nacre or bluish in color. The content of the mucocele is white-greyish, white-yellowish or brown.

Mucus is a complex secretion synthesized and secreted by specialized goblet and mucous cells in the columnar epithelia that line the lumen of all of the organs and glands that are exposed to and communicate with the external environment [10] having a protective function for the underlying epithelia [11]. The data presently available indicates that fluid secretion by these glands is mainly under muscarinic control and is due to acinar Cl-secretion driven by a basolateral Na+-K+-2Cl⁻ cotransporter.

Mucus has been described in organisms from all kingdoms [12] as being a complex dilute aqueous viscoelastic secretion consisting of following components: water, lipids, various proteins and electrolytes [13].

Histologically, paranasal sinus mucoceles have respiratory mucous characteristics and the walls of the mucocele are made from pseudostratified ciliated columnar epithelium [12,14,15]. It is rare for a mucocele to present with metaplasia but there are chronic cases that can present squamous metaplasia [10]. In the cystic epithelium reagents may form [16]. In the mucocele, there is an increased IL12 expression that leads to an increase in interferon gamma. Consequently, activation of T lymphocyte accelerates the occurrence of chronic inflammatory infiltrate [14,17]. There is also cholesterol granuloma, fibrosis, granulation tissue and local bleeding.

Concerning the biochemical analysis performed in the 10 cases: no relationship was found between the concentration of the elements of the mucocele fluid and their evolution and no significant variation in their concentration.

Electrolytes are an important component of mucus and their composition can be different in mucus from various tissues, function on the underlying secretory epithelium. These differences may be important to the specific environmental challenges found in each organ. Common to most mucus secretions are sodium and potassium chloride and sodium bicarbonate, phosphate and magnesium and calcium, approximately isotonic with serum. Some organs modify the ionic composition of mucus during their specialized functions or during disease. Physiologically, changes in the electrolyte composition have a role in controlling mucus hydration and rheology, *e.g.* increase in concentration of monovalent cations, Na or K can cause a reduction in mucus viscosity [18]. Normal airway mucus hydration (and viscoelasticity) is regulated by at least two signaling systems mediated by ATP and adenosine. To hydrate airway mucus, the adenosine system normally inhibits Na⁺ reabsorption and initiates Cl secretion. In different diseases these functions cannot be performed, Na⁺ reabsorption causes water reabsorption by osmosis, leading to mucus dehydration, flattened cilia, and stasis [19].

In our study group we found modified electrolytes values compared to normal findings and this could lead to the assumption that mucous composition is modified according to a balance or unbalance between at least these two systems (ATP and adenosine).

From the clinical point of view, the following forms can be encountered:

Mucocele in the frontal sinus

- *the ethmoid-fronto-orbital form* - we find it most frequently. It is manifested by: swelling in the super-internal angle of the orbit, eye signs - exophthalmia (ocular globe deflected down and out) and a decrease in visual acuity; frequent recurrent disease is associated with this localization.

- *pure orbital* mucocele is the ophthalmic form; it is characterized by an isolated exophthalmia in the absence of visible exteriorization; E.N.T. is not regularly consulted and therefore diagnostic error is frequent.

- *frontal* mucocele - external orbits - rare forms - occur when the mucous membrane develops in the external extension of a large frontal sinus.

- *pure frontal* mucocele - is exceptional. This form of mucocele is characterized by a frontal swelling, which translates the presence of the mucocele into the frontal sinus.

Mucocele of the ethmoid

a. Anterior mucoceles, which after the predominant extension has the following forms:

- the orbital form characterized by: swelling on the inner wall of the orbit and eye signs: exophthalmia (ocular globe deflected outside), diplopia and epiphysis;

- endonasal form characterized by: endonasal swelling; unilateral nasal obstruction and headaches;

- Mixed form is the most common in encountering nasal and orbital symptoms;

- the pure ethmoid form is relatively rare;

- the mucocele of the bulla determines: narrowing the middle meatus and compressing the middle turbinate through the ectasia of the bulla).

- the middle turbinate mucocele is the most common of the localized ethmoid mucocele. It is characterized by: a very large middle turbinate that pushes the nasal septum and can touch the nasal floor and vestibule, nasal obstruction, anosmia, headache.

b. Posterior mucoceles: arise from the cells of the posterior ethmoid (exceptional).

The sphenoid sinus mucocele is manifested by a symptomatic triad with three main ophthalmic signs (drooping of the eyelid, diplopia and restricted ocular movements).

Maxillary sinus mucocele

- the ethmoid - maxillary form;

pure maxillary sinus mucocele; is characterized by the deformation of the jugal region (canine fossa).

In the literature, the positive mucocele diagnosis is considered easy, provided you think about it.

Generally, the anterior mucocele, which is the most common, allows for a positive diagnosis. The posterior mucoceles, especially in the sphenoid sinus, are diagnosed with difficulty and in advanced stages because they do not manifest themselves clinically unless, by their size, they cause mechanical complications on the neighboring regions.

The predisposing factors that could have a role in mucocel development are cranial: cranial dysplasia, fibroosseous lesions, chronic rhinosinusitis, facial fractures or rhinonasal manifestations of systemic diseases [4,17].

Among the cases we had studied, the majority were 24 frontoethmoid mucoceles.

The pain and rhinological symptomatology pointed for the diagnose and the indication of a CT scan confirmed the diagnosis. In three cases the mucocele was located in the maxillary sinus and there were only 2 cases associated with pain. One of the cases did not show symptoms-known as the *silent sinus* syndrome [20].

In all cases the diagnosis was confirmed after the CT examination. We present some aspects of our cases (fig. 1-5).

Diagnosis is confirmed by a CT scan, which reveals an expansive soft tissue lesion stretching the surrounding bone that appears thinner.

The CT scan provides the basic anatomical aspects of the mucocele, separating it from neighboring bone and aids in establishing a treatment plan [14, 21]. It has also good value in differential diagnosis with different anatomic variations [22]. CBCT scans are also an option in diagnostic, with good detail regarding maxilla bone and paranasal sinusal architecture. Complex tridimensional reconstructions allow correct evaluation and optimal treatment strategy [23, 27, 28].

An MRI scan shows the relationship of the mucocele with its surrounding soft structures. The intensity of the signal in T1 and T2 is dependent on the viscosity of the fluid contained in the mucocele [5]. In T2 the mucocele is hyperintense due to increased water content. Over time this intensity decreases. In contrast, T1 has low signal strength due to water resonance and an increased protein concentration.

The viscosity of the mucocele changes from an isointensive structure to a hyperintense structure [16, 24,29,30].

Depending on the location, the mucocele determines the symptoms that can be grouped into: rhinological, ophthalmological and neurological.

rhinological symptoms - when the rhinosinusal region is affected and are characterized by a pressure sensation, soreness in the affected sinus, obstructive nasal syndrome, usually unilateral, sometimes rhinorrhea,

ophthalmic symptoms when the growth develops toward the orbital region and palpebral plexus, periorbital swelling, exophthalmia, ocular pain, vision changes, opthalmoplegia occur;

- neurological symptoms - manifested by headache.

Regarding the patients included in the study, 12 patients presented with a secondary mucocele: 8 patients suffered a traumatic event prior to the diagnosis and 4 patients had a history of surgery in this area. We have noticed that in the



maxillary sinus etmoidal sinus

Fig. 2b. Mucocele in the right etmoidal sinus

Fig 3. Mucocele in the left ethmoid sinus, with lateralization of the lamina papiraceea



Fig. 4. Mucocele in the right fronto-ethmoidal recess



Fig. 5. Left frontal sinus mucocele

case of patients with a history of trauma, the time passed between the accident and the time when the symptoms pointed towards the diagnosis of mucocele varied between 10 and 16 years.

In the case of patients who underwent FESS interventions, the time between surgery for previous pain and the recurrence of the mucocele was much shorter, ranging about 1.5-2 years. This was also observed by other authors [3,10,31-33].

The time difference between the time of diagnosis between those who underwent FESS-type interventions and those who have had trauma can be linked to mucosal inflammation following the FESS intervention that may accelerate the development of the mucocele.

It is therefore necessary to follow-up patients who have had interventions for an extended period of time [3,34-36].

Also, given that mucosal trauma appears to be an important factor in mucocele formation, surgical trauma should be minimized as much as possible. These patients will be regularly monitored endoscopically to avoid the risk of mucocele recurrence [3,25,37,38]. In all of our cases, the treatment was a surgical external approach in order to remove the mucocele capsule and ensure large drainage to prevent recurrence.

We considered that early intervention is indicated to prevent mucocele enlargement in areas of importance that may affect prognosis or delay healing. The current trend is to approach the mucocele by endoscopic techniques which reduces the length of hospitalization and the number of recurrences [5, 26,39,40]. Patients were followed-up at one month post-operatively and then at 3 months, 6 months and 1 year.

Conclusions

Mucoceles can be discovered late, when associated signs like ophtalmological simptoms appear visible but they have a high rate of healing after effective surgical approach.

Trauma or rhino-sinusal surgery can create the opportunity for mucoceles formation. If the predisposing factor is rhinosinusal surgery, mucoceles have the tendency to develop symptoms more quickly than in primary mucoceles.

Analysis of the components in the sinus fluid may provide new indications in the mechanism of mucocele formation.

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