MUCOCELES

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Introduction

ALTHOUGH otorhinolaryngological literature contains many cases of mucoceles since the condition was first reported in 1839, little has been written in recent years concerning the possible mechanism and pathology of this condition.

Since it has been our experience to see four cases during the past nine months in which a diagnosis of mucocele was made, we should like to very briefly report these cases, discuss their diagnosis and treatment, and on the basis of careful pathological studies present what we believe is a more understandable concept of the mechanism of mucocele formation.

Case No. 1

A 66-year-old white male was admitted to the Illinois Eye and Ear Infirmary on September 10th, 1947, with the following history: four years before he noted a small, painless swelling over the left eye, which became progressively larger causing a marked facial deformity, diminished vision, and a downward and outward displacement of the eye. No history of trauma or infection could be elicited and the past history for rhinological difficulties was non-contributory.

Ophthalmological findings consisted of: a cystic like swelling above and in the left orbit extending forward approximately one and one-half inches causing a downward and outward displacement of the eye. Vision was recorded as 20/200 in the left eye. There was no diplopia.

Otorhinolaryngological examination revealed no unusual findings. X-ray films of the skull revealed a destructive process involving the superior and medial portion of the left orbital ridge, the frontal sinus septum and upper medial angle of the right orbital ridge. The left anterior ethmoids appeared to be partially destroyed. On September 11th, a diagnostic aspiration of the cystic mass was made with about 5 c.c. of a brownish tenacious sero-sangineous fluid being obtained. On the basis of the eye changes, diagnostic aspiration, and X-ray findings a diagnosis of mucocele was made.

On September 12th, an external frontal operation was performed and the mucocele removed. The floor of the frontal sinus, the medial wall, and the interfrontal septum had been completely destroyed. The posterior and lower wall of the frontal sinus had been eroded and an

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extradural abscess 1½ cm. in circumference had formed. All possible necrotic debris was removed and the entire ethmoid labyrinth exenterated. A large drain was placed from the sinus into the nose and the skin closed.

The patient's postoperative course was uneventful.

Case No. 2

A 25-year-old negro male presented himself at the Illinois Eye and Ear Infirmary on April 28th, 1947, because he had noticed a drooping of the left eyelid during the past year.

He stated that in 1936 he sustained a fall on the ice cutting the skin over the left eyebrow, which required five stitches to close. In 1938, he fell off a street car and injured the left side of his head, but was not seriously incapacitated. No history of past rhinological difficulties, headaches, diplopia, or vertigo could be obtained.

Ophthalmological findings consisted of a sensation of fullness of the inner canthus of the left eye and vision of 20/40 for this eye. The remainder of their examination was normal except for a slight ptosis of the left lid and a downward and outward displacement of the left eye.

Otorhinolaryngological examination revealed no abnormal findings and the rhinoscopic picture in particular was normal. X-ray studies of the frontal sinuses revealed a depression of the superior margin of the left orbit consistent with a mucocele.

An external frontal operation was performed under local anæsthesia and the mucocele of the left frontal sinus was removed in its entirety, together with the entire mucous membrane and necrotic bone of the sinus. The naso-frontal ostium was enlarged and a rubber tube was placed from the sinus through the nose, after the anterior one-third of the middle turbinate had been removed.

The patient's postoperative course was uneventful.

Case No. 3

A 43-year-old negress presented herself at the Illinois Research Hospital on October 1st, 1947, because of a painfully protruding eyeball of six months' duration.

She stated that in April 1947 she had a severe cold followed by sinus trouble after which she noticed that her left eyeball began to protrude, her vision began to be impaired, and her eye motility limited.

Ophthalmological findings consisted of: a proptosis of the left eye to 25 mm. in a downward and outward direction with marked engorgement of the retinal veins. Disc margins were hazy suggesting œdema of the disc. Vision was recorded as 20/25 plus 3.

Otorhinolaryngological examination revealed a septal deviation to

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the left with œdematous mucosa of the turbinates but no pathological secretions. X-rays revealed destruction of the superior lateral margin of the left frontal sinus involving the roof of the left orbit. A roentgen diagnosis of osteomyelitis with bony destruction of the superior and lateral walls of the left frontal sinus and of the roof of the left orbit with left ethmoiditis was made. These findings together with the eye changes suggested a mucocele or pyocele of the frontal sinus.

On October 17th, 1947, a submucous resection with the removal of the anterior one-third of the middle turbinate was performed. Two weeks later an external frontal operation was done but no mucocele was found. The medial aspect of the sinus was soft and tissue was removed for biopsy. The naso-frontal ostium was enlarged and a rubber catheter sutured in place. The skin was closed.

The pathologist reported the biopsy material as Carcinoma.

The patient had a foul discharge from the wound postoperatively and by mid-January 1948 had become bed-ridden because of weakness and pain. On January 23rd, 1948, she was re-hospitalized and a radical exenteration of the left orbit with removal of a large part of the left frontal and ethmoid sinuses together with the zygoma and the great wing of the sphenoid was performed. The area was extensively involved by carcinoma with apparent extension to the dura. The wound was left open for postoperative X-ray therapy.

To date the patient has received extensive postoperative X-ray therapy and is being followed regularly in the Tumour Clinic.

Case No. 4

A 51-year-old white male presented himself at the Illinois Eye and Ear Infirmary on February 13th, 1948, because of pain over the right frontal and maxillary sinuses and dryness of the throat.

The otorhinolaryngological examination revealed a marked injection of the mucous membrane of the nose and pharynx. X-rays of the sinuses revealed a large rounded mass of slight increase in density within the right antrum with slight haziness of the right frontal sinus. A roentgen diagnosis of mucocele of the right antrum and a mild right frontal sinusitis was made. Lipiodol studies of the right antrum confirmed the diagnosis of mucocele.

On February 27th, 1948, a Caldwell-Luc was performed on the right antrum and the large mucocele was removed and sent to the laboratory for biopsy study. The patient's postoperative course was uneventful.

Diagnosis

From these cases it would appear that the diagnosis of a mucocele in the advanced case is simple, in the complicated, confusing, and in the early, extremely difficult.

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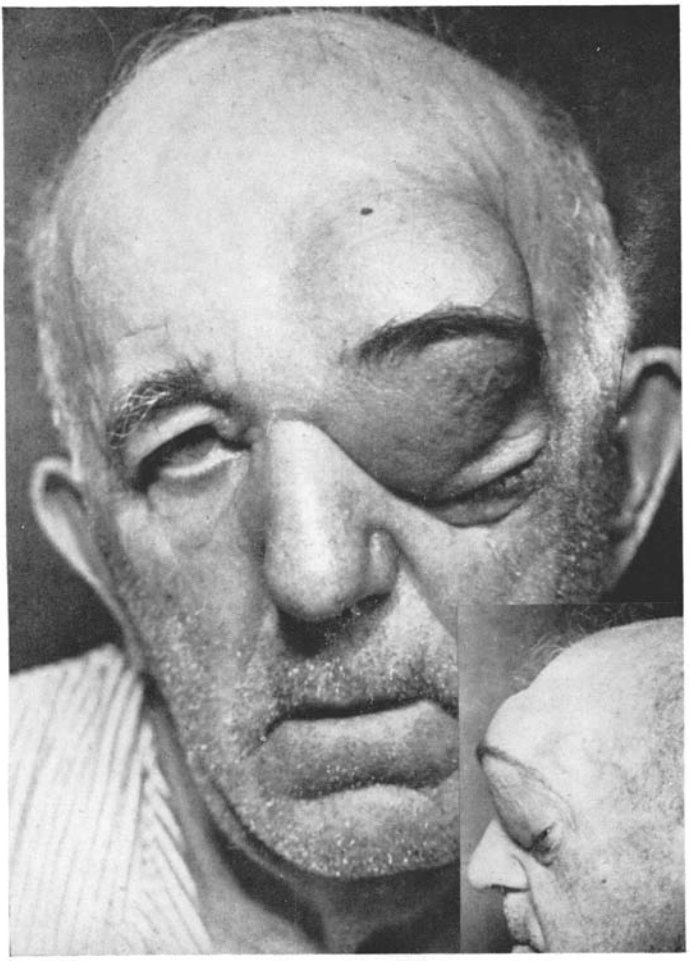


FIG. 1.
Showing front and side view before surgery.

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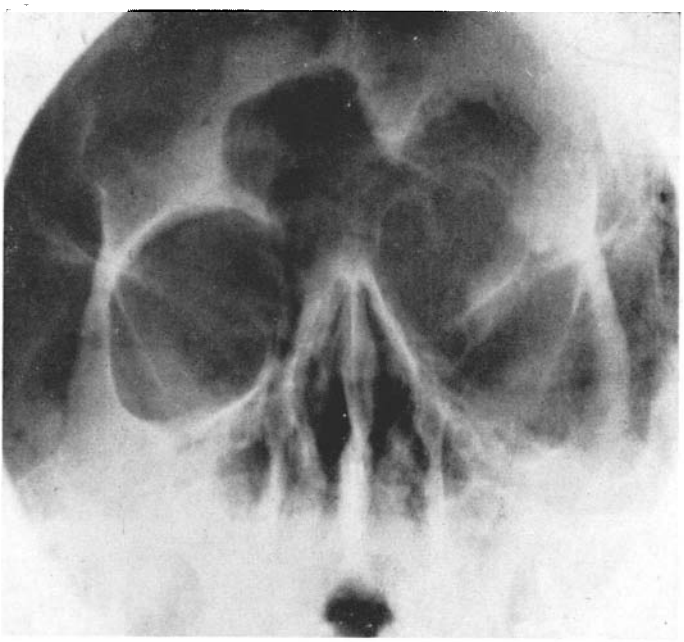


FIG. 2.
X-ray showing destruction of the frontal and ethmoidal sinus.

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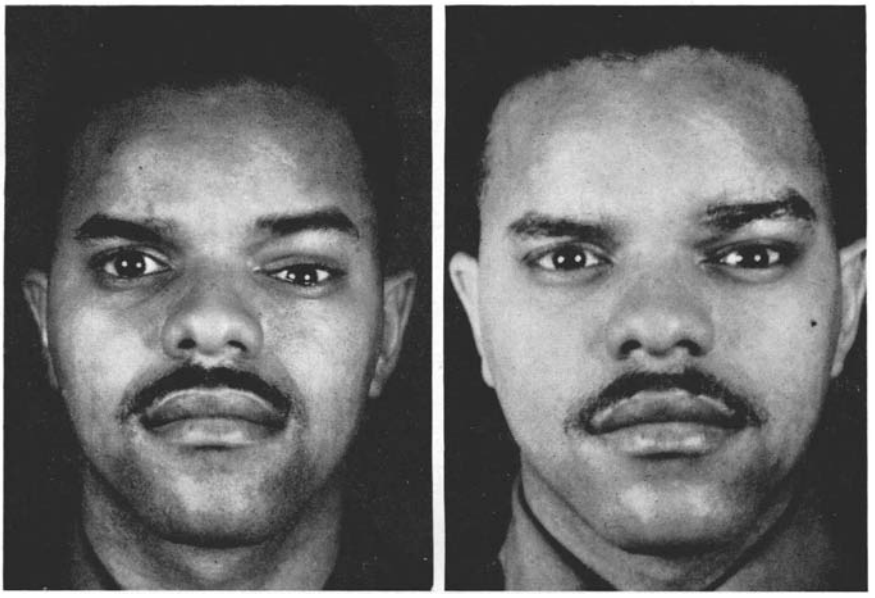


FIG. 3.
Showing front view before and after surgery.



FIG. 4.
Thickened wall of a mucocele with epithelial lining outwards.

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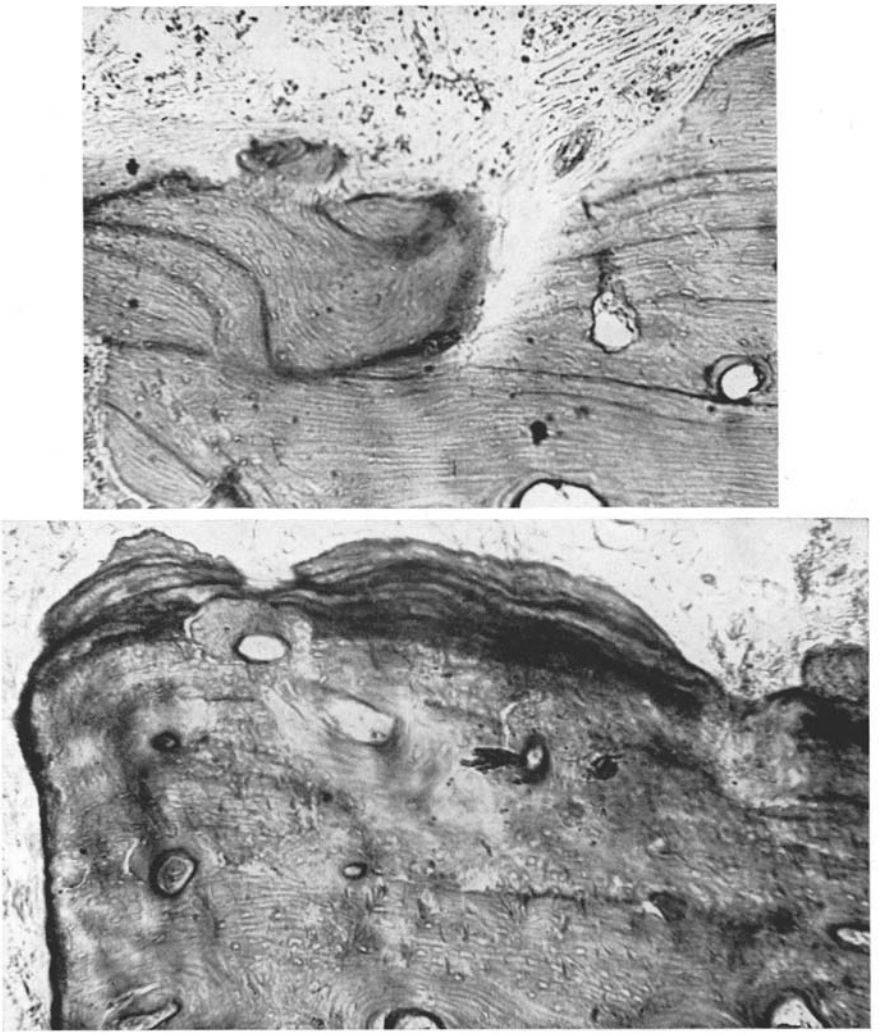


FIG. 5.
Bony changes of the sinus wall with pressure necrosis and bone apposition.

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Mucoceles

In most cases the patient is first seen by the ophthalmologist because of visual complaints and secondarily by the otolaryngologist.

The history generally given by the patient is one of diminished vision, diplopia, and a painless progressive displacement of the eye in a downward and outward direction. On further inquiry he may recall an injury to the head a number of years previously, or recount a history of a severe sinus infection in the past.

The otolaryngologist is presented with a case which must be differentiated from: gummas, aneurysm, fibromata, angiomata, lymphoid tumours, meningoceles, carcinoma, osteoma, and xanthomatosis. A case with all of the classic findings of a mucocele has been reported in the literature which proved to be a case of xanthomatosis.

The otorhinolaryngological examination is very often unrevealing and the rhinoscopic examination in particular is generally normal. The otolaryngologist's suspicion may be aroused by a parchment-like crackling due to the partial absorption of the bone. His chief armamentarium, however, consists in a diagnostic aspiration, which if positive for thick brownish tenacious fluid is presumptive evidence of mucocele, and the X-ray, which shows destruction of bone, and in the absence of other findings can be interpreted as additional evidence of mucocele. Even after a tentative diagnosis of mucocele has been made, measures should be taken to insure an immediate frozen-section pathological study at the time of surgery in the event that a malignancy is encountered.

Treatment

On the basis of the operative findings in the cases of this series together with the extensive pathological changes to be mentioned below, it seems to us that the only adequate therapeutic measure lies in a radical exenteration through an external incision combined with the establishment of adequate intranasal drainage. Intranasal procedures alone as advocated by Goodyear are inadequate except when radiological findings are negative and the bone is intact.

The intranasal procedure has been found to be temporizing with a serious condition and in two cases observed by us has led to a recurrence of the mucocele.

The external operation should be made through the Killian incision for mucoceles of the frontal sinus. The technique advised by Lederer of making the incision through the line of the eyebrow growth, which is clipped preoperatively, has been found to enhance the postoperative appearance of the patient, making the line of incision almost imperceptible. Following the removal of the mucocele from the frontal sinus, the cavity should be radically exenterated of all suspicious membrane and the ethmoid sinuses should be thoroughly curetted.

The external operation for mucoceles of the maxillary sinus should

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consist of a well-executed Caldwell-Luc procedure. The formation of an antral window in surgery for this condition is optional.

The Mechanism of Mucocele

At the present time the mechanism of mucocele formation is believed to be a cystic accumulation of fluid, resulting from either trauma or infection, which undergoes active enlargement within a sinus causing atrophy of the surrounding bony structure. On the basis of a careful study of sections obtained from the four cases presented above we should like to advance another concept for the explanation of mucocele formation.

Pathological specimens, upon which this new concept is based, lead us to believe that initially there is a submucosal injury, either hæmorrhage caused by trauma or as a result of an invading infection, which causes a localized elevation of the mucosa. It will be apparent that, in contrast to the other theories, this elevated mucosa forms the beginning wall of the future mucocele and by the herniation of the mucosa toward the cavity of the sinus as gradual development occurs by the accumulation of its contents, the wall of the mucocele is going to consist outwardly of the pseudostratified columnar ciliated epithelium and inwardly of fibrous connective tissue which has been stripped away from the endosteum. Because of the resistance offered by the sinus mucosa to this submucosal elevation, the progress and development of the mucocele is a rather slow one. When the elevated and herniated mucosa finally touches the intact sinus mucosa of one of the surrounding sinus walls and the submucosal distension persists pressure is exerted upon both the mucosa and the underlying bone. This explains the history of trauma sustained many years previously which is elicited from patients who have the characteristic visual signs and symptoms of mucocele. Indeed the trauma may have occurred so long before that the patient cannot recall the incident.

To recapitulate, the main points in the mechanism of mucocele formation seem to be: (1) A submucosal injury either hæmorrhagic, traumatic, or infectious; (2) an elevation of the mucosa caused by an accumulation of exudate beneath it; (3) a herniation of the elevated mucosa toward the centre of the sinus cavity; and (4) pressure of the herniated mass upon normal sinus mucosa and the underlying bone.

The Pathology of Mucoceles

The histo-pathology of mucoceles as revealed by the specimens of our series may be considered under three parts: (1) Contents; (2) Mucosal changes; and (3) Bone changes.

1. The contents of the mucoceles consist chiefly of old blood elements, accumulations of red blood cells, red cells suspended in serous fluid,

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fibrous elements, and large cells. Significantly, there are no cholesterol crystals present. It is known that cholesterol crystals are found in cysts derived from ectoderm such as dental cysts, but are absent from growths arising from mesoderm and are thus absent in mucoceles.

2. The external mucosal changes consist primarily in a modification of the normal sinus mucosa. The normal cilia are absent and the columnar cells become cuboidal in type as a result of the pressure exerted by the developing mucocele.

The inner wall of the mucocele is considerably thicker at the base where the initial submucosal injury resulted in an elevation and herniation of the sinus mucosa. It becomes thinner as the mucocele enlarges and contacts the osseous walls of the sinus. Microscopically the wall consists of an internal fibrous layer and an external layer of epithelium with the changes mentioned above.

Grossly, the mucocele appears bluish if the contents are brown and pale if the contents are colourless.

3. The changes of the adjacent bone are those characteristic of bones influenced by constant pressure and tension. In the beginning of the process, the pressure stimulates bone apposition and leads later, with an increase of pressure, to bone resorption which prevails above the first changes, giving the bone a thin parchment-like appearance.

In microscopic sections one can see the endosteum replaced by a loose fibrous membrane infiltrated with necrotic bone cells and blood cells. The surface of the bone shows areas of recent absorption with osteoclastic activity and apposition of a new immature bone. The marrow tissue spaces in the deeper areas are transformed to fibrous tissue cavities with an active bone absorption.

The external bony lamina appears to be affected by the pathologic changes of the inner surface. One can see areas of a very recent reconstruction with active bone apposition under the thickened periosteum. The circumferential lamellae underneath the periosteum show a dense area of reversal and resting lines interrupted by a few old Haversian systems. The marrow tissue spaces and Haversian canals are enlarged and partly filled with fibrous elements. The lacunae of the lamella are empty because of the absence of the osteocytes.

Signs of inflammation and cellular infiltration are not visible and the entire process seems to be the result of a metabolic and physical alteration rather than one of an inflammatory osteitis.

Summary and Conclusion

We have presented four cases in which a diagnosis of mucocele had been made in our clinics within a nine month period. We have reviewed very briefly the symptomatology, the problems of diagnosis, and the manner in which we feel such conditions should be adequately treated.

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On the evidence gained from a careful pathological study of the material from these cases, we feel that a mucocele is not due either to cyst formation in the lining membrane of the sinus or to the blocking of the sinus ostium. On the strength of this evidence we have presented what we believe to be a new concept in the mechanism of mucocele formation, namely: (1) a submucosal injury occurs as a result of traumatic hæmorrhage or infection, resulting in (2) an elevation of the mucosa with an accumulation of exudate beneath it; and that (3) a herniation of this elevated mucosa occurs until it encounters the resistance of normal sinus mucosa of one of the sinus walls where (4) pressure is exerted upon the mucosa and the underlying bone producing characteristic changes.

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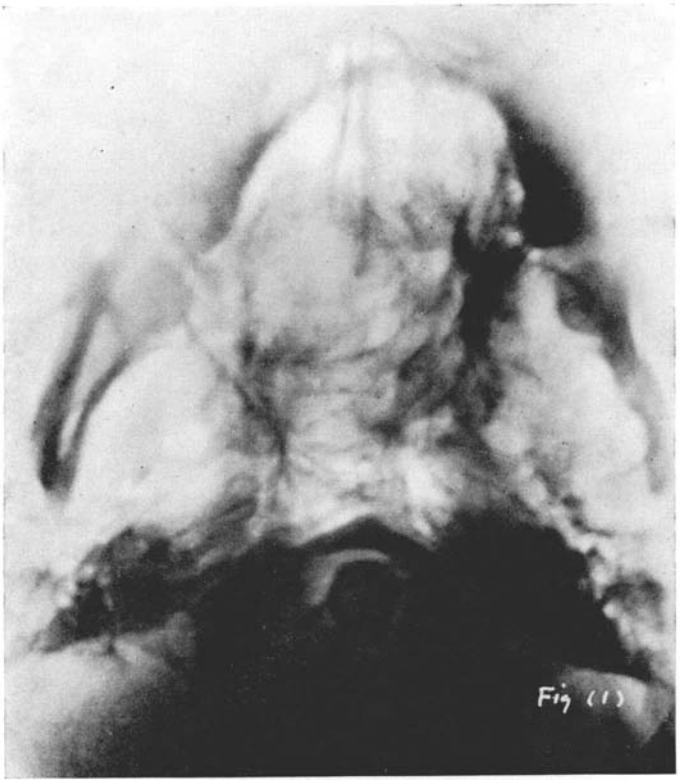


Fig. 1.

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FIG. 2.

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