



“Internal decompressive craniectomy with craniotomy: a novel surgical therapy of giant frontal mucocele complicated by subdural empyema

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Abstract

Introduction: Giant frontal mucocele (GFM) is an extremely rare cause of frontal lobe syndrome. Subdural empyema (SDE) is an uncommon complication of paranasal sinusitis, for which craniotomy and decompressive craniectomy are the most effective surgical procedures.

*Case report: A 54-year-old man was brought unconscious to the Emergency Room where recurrent generalized seizures occurred. Heroin abuse, HCV related hepatitis, prolonged antibiotic therapy for treatment of purulent rhinorrhea, along with recent personality changes, was reported. High white blood cell count, pansinusitis, GFM, SDE and cerebritis were documented. The patient underwent bifrontal craniotomy in emergency, extensive drilling of the inner aspect of the frontal bone, surgical toilette of the enlarged frontal sinus and its “cranialization”. *Prevotella intermedia* and *Fusobacterium nucleatum* were isolated and antibiotic therapy was started intravenously and then continued orally for three months. Two years later the patient has recovered, though minor signs of frontal lobe syndrome persist.*

Discussion: To the Authors knowledge this is the first case of GFM with SDE reported in the literature. Although decompressive craniectomy is advocated in extreme conditions, as in this case, “internal decompressive craniectomy”, obtained with craniotomy and cranialization of the frontal sinuses, is strongly advocated in cases of SDE associated with megasinuses.

Key words: Mucocele; subdural empyema; internal decompressive craniectomy.

Introduction

A mucocele is an epithelium lined mucus containing sac, which usually develops when the ostium of paranasal sinus becomes obstructed by chronic sinusitis, polyps or bone tumours. Impediments to sinus ostium ventilation are thought to be the princi-

pal cause of formation, resulting from anatomical obstruction, mucosal hyperplasia, mass lesion or other mechanical factors. Mucoceles can erode through the surrounding bone and spread intracranially (1). Surgical treatments include endoscopic sinus surgery and craniofacial approach with or without sinus obliteration (2, 3). Mucocele invasion of the orbit and anterior cranial fossa requires an aggressive approach to avoid ophthalmologic or neurological complication. In this paper, we report a case of a giant fronto-ethmoidal mucocele extending into the intracranial region and producing a subdural fluid collection consistent with empyema, associated with mass effect and overlying soft tissue swelling secondary to cerebritis. “Internal decompressive craniectomy with craniotomy”, never described as alternative procedure to the classic decompressive craniectomy, was performed along with prolonged antibiotic therapy.

Case report

A 54-year-old man was brought unconscious to the Emergency Room, where recurrent generalized seizures occurred. The patient presented with insidious onset of a progressive enlarging bony, cosmetically unacceptable, forehead swelling without proptosis. Injection drug abuse (heroin), HCV related hepatitis, prolonged antibiotic therapy for the treatment of a purulent rhinorrhea in mucocele, along with recent personality changes, were reported.

A high white blood cell count was consistent with infection (white blood cell count was 22.000/ microl with 26% band forms, 48% segmented neutrophils and 10% lymphocytes).

MR Imaging and CT scans documented extensive pansinusitis along with a giant frontal sinus

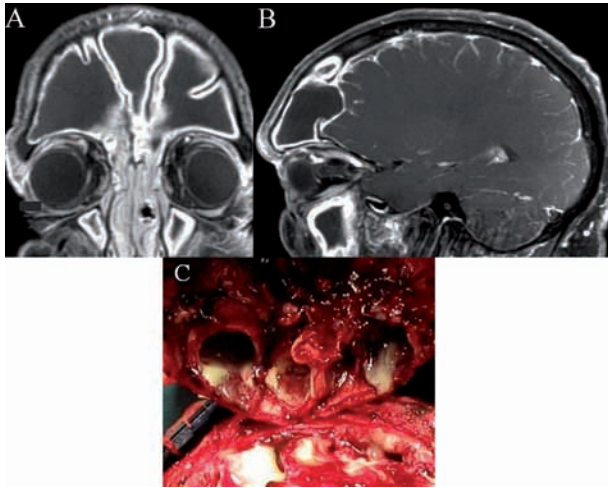


FIG. 1. — A-B: Coronal and sagittal post-contrast T1-weighted images documenting the frontal mucocoele, that has eroded the posterior wall of the frontal sinus, with a resulting subdural empyema and signs of cerebritis. C: Intraoperative view of the giant frontal sinus filled with pus.

mucocoele (GFM), subdural fluid collection consistent with empyema (SDE), associated with mass effect and overlying soft tissue swelling consistent with cerebritis (Fig. 1A-B). The patient underwent a bifrontal craniotomy in emergency with transverse opening of GFM bilaterally (Fig. 1C). A huge breakthrough on the posterior wall of the frontal sinuses was evident bilaterally (left more than right site) along with bilateral subdural empyema and cerebritis, more consistent on the left site. Suction of the fluid collection, surgical removal of the debris, local irrigation with saline and wide spectrum antibiotic (Ceftriazone 2 mg in 100 saline) was performed in the subdural space. A bifrontal cerebral herniation due to the cerebritis and brain swelling was evident. Therefore a complete removal of the posterior wall of the giant frontal sinuses by using high-speed pneumatic drill and meticulous surgical remodelling of the inner table of the frontal bone were performed; this procedure permitted to gain at least 1 cm frontally. Removal of the flogistic sinus mucosa, local bipolar cauterization, sinus filling with gelfoam and antibiotics (Ceftriazone 2 mg in 100 saline) along with its “cranialization” with wide dural patch and muscle, Gelfoam and Fibrin glue were performed on the frontal sinus. Intravenously wide spectrum antibiotics were administered intraoperatively (Ceftriazone 2 mg).

Gram stain of the SDE showed many polymorphonuclear white blood cells along with two different microbic agents: *Prevotella intermedia* and *Fusobacterium nucleatum*. HIV seroconversion testing was negative. Etiologic antibiotic therapy with

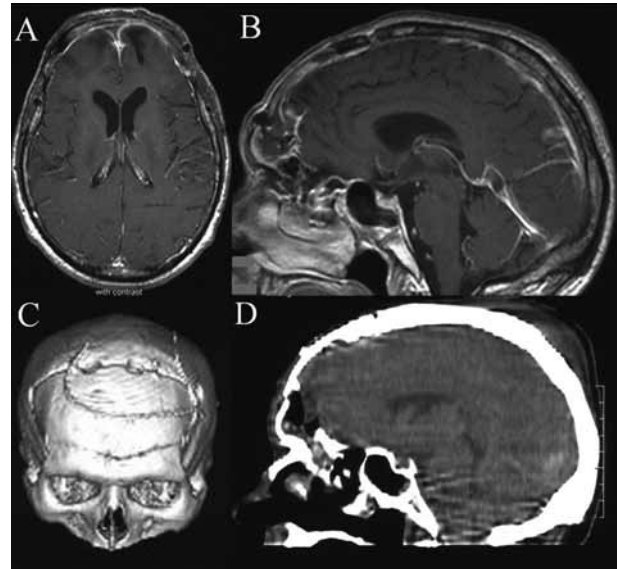


FIG. 2. — A, B: Post-operative axial and sagittal post-contrast T1-weighted images documenting no disease recurrence and the frontal lobes injury sequelae. C, D: Immediate postoperative axial CT scan with 3D reconstructions showing the profiles and key holes of the internal decompressive craniectomy with craniotomy along with the brain swelling adaptation to the new internal space.

amoxicillin and clavulanate potassium (6 g/day) along with metronidazole (2 g/day) was administered intravenously two days after surgery for 7 days and then continued orally for 3 months. The patient's neurological conditions immediately improved after the surgical procedure and he was discharged with antiepileptic drug therapy (Phenobarbital 150 mg daily). Post-operative MR Images showed the absence of disease recurrence and frontal lobes injury sequelae (Fig. 2A-B), sagittal CT scans with 3D reconstructions demonstrated the profiles and key holes of the internal decompressive craniectomy with craniotomy along with the brain swelling adaptation to the new internal space (Fig. 2C-D).

Two years later the patient has recovered, though minor signs of frontal lobe syndrome persist. Although no seizures occurred in the last two years the patient continued antiepileptic drug with lower dosage (Phenobarbital 100 mg daily) and then progressively discontinued. At the latest follow up the patient is seizures free.

Discussion

We will focus on three topics in the discussion of this case report: the GFM, the SDE and the surgical therapy of GFM complicated by SDE.

GIANT FRONTAL MUCOCELE

Rollet first used the term mucocele in 1896 (4). The GFM is a benign entity caused by retention of mucous secretions in the frontal sinus. Among the facial sinuses it is the most frequent compared to the ethmoidal, sphenoidal and maxillary; it usually occurs during the third and fourth decades of life and its incidence is similar for both genders (4). Accumulation and retention of mucous secretions in a sinus can be caused by different factors. For this reason primary or secondary paranasal sinus mucoceles have been described. For the formation of primary mucoceles, inflammatory blockage of mucus drainage, secretory duct obstruction, cystic dilation of mucosal glands, and cystic degeneration of polyps were proposed to be the possible mechanism (5, 6). For the secondary mucoceles, sequestering of residual mucosa in the wound and long-term retention of tissue fluid were advocated to lead to their formation (6, 7). Therefore our case can be described as a primary mucocele. Primary or secondary sinus mucoceles have different patterns of aggressiveness regarding orbital or cranial extension (8).

GFM may expand and erode the surrounding structure such as bones and cerebral parenchyma, causing intense headache (9). More rarely GFM can cause frontal lobe syndrome (10) and transient motor dysfunction. Fu *et al.* reported 3 cases with intracranial extension out of 19 cases of primary frontal mucoceles, but no cases of intracranial extension in 16 cases of secondary frontal mucoceles (8). An associated subdural empyema has never been described.

Usually GFM invades intracranial spaces eroding the posterior wall of the frontal sinus, causing meningitis, meningoencephalitis, pneumocephalus, brain abscess, seizures or CSF fistula (11). On the other hand, intracranial extension of frontal sinusitis is often not a direct spread through frontal sinus' posterior wall erosion, but it is more commonly a retrograde thrombophlebitis via the valveless diploe veins, facilitated by the shared venous drainage of sinuses and intracranial structures (12). The diagnosis of mucocele is based on the history, physical examination and radiological findings. CT scans and MR Images are effective in detecting the lesion and in demonstrating any intracranial extension. Mucoceles can be hyperdense or hypodense on CT, although brain and mucocele densities are usually equal. On T1-weighted MRI, the lesion can appear either homogeneous or heterogeneous, and different signal patterns (such as hyper-, hypo-, and isointensity) can be appreciated as well (9, 13, 14).

Surgical treatment of GFM includes excision, skull base dural plastic and wide opening of the frontal sinuses by using intranasal endoscopic approach and/or frontal craniotomy (15). Some surgeons advocate two stages surgical therapy to avoid infections and facilitate radical excision of the mucoceles, otherwise single stage surgery has been more commonly recommended due to the availability of antibiotics and fibrin glue sealants (4, 16).

SUBDURAL EMPYEMA

A suppurative intracranial infection such as SDE is an uncommon potential life threatening complication of sinusitis, representing fewer than 10% of the intracranial complications (12, 17, 18, 19).

The frontal sinus is the most common sinus associated with intracranial infection, followed by the ethmoidal, sphenoidal, and maxillary sinuses (20). The maxillary, ethmoid, sphenoid, and frontal sinuses all share thin bony walls with the orbit and the cranium.

The SDE is a surgical emergency. Although SDEs as intracranial complications of sinusitis are rare, the morbidity and mortality remain high. It is therefore necessary to diagnose and treat intracranial infections promptly. A SDE usually presents as an insidious process. Early symptoms are usually non-specific. The most common presenting clinical features are indirect and dealing with purulent rhinorrhea, headache and fever. Patients with frontal sinusitis demonstrating persistent fever, forehead swelling or failure of resolution of symptoms warrant radiological evaluation to look for silent intracranial involvement, even in the absence of intracranial signs (21). Unless diagnosed early, the condition can progress rapidly, leading to neurological deterioration due to the infection and to the secondary intracranial hypertension. The delay in diagnosis is mainly due to the non-specific initial presenting symptoms.

CT scan is the diagnostic imaging modality of choice. It confirms the presence and delineates the extent of the intracranial involvement. The classic appearance of SDE on a CT scan is a thin low-density collection over the cerebral convexity or in the interhemispheric fissure that has a rim of contrast enhancement (22, 23, 24). A CT scan with contrast is usually sufficiently sensitive to detect a SDE. In cases where the clinical condition suggests such complications, a negative CT scan does not rule them out and MRI should be obtained (25).

The cultures from frontal sinusitis with complications frequently reveal polymicrobial involvement. Streptococci, staphylococci and anaerobic bacteria are the predominant pathogens. Anaerobic strepto-

cocci have been shown to be the most common microorganisms isolated in SDE (12, 26, 27, 28). On the contrary, *Prevotella intermedia* and *Fusobacterium nucleatum* (both involved in odontal and parodontal pathologies) were isolated in our case (29, 30, 31, 32, 33, 34).

Recurring subdural empyema, despite adequate surgical drainage and antibiotic treatment, is still a life threatening disease; this is mainly due to poor diffusion of the antibiotics into the subdural space (35). In our opinion a radical and effective medical and surgical treatment is strongly advised in "one staged procedure".

SURGICAL THERAPY: THE ROLE OF INTERNAL DECOMPRESSIVE CRANIECTOMY WITH CRANIOTOMY

Several treatment options are available and the choice depends on the extension degree. Surgical treatments of frontal mucocèles include endoscopic sinus surgery and craniofacial approach with or without sinus obliteration (2, 3). Mucocèle invasion of the orbit and anterior cranial fossa requires an aggressive approach to eradicating the lesion. Some authors believe that an intranasal approach is the first choice, even in patients with intracranial extension (36). They argue that this approach is less invasive and they emphasize the possibility that the lesion can be diagnosed and completely removed without craniotomy. On the other hand, Delfini et al prefer a transcranial approach for anterior mucocèles with intracranial extension to ensure complete removal and to prevent recurrence (37).

In our opinion, intranasal approach can be used when the lesion is confined to the paranasal sinuses. In cases of primary giant frontal mucocèles, the risk of intracranial involvement is high, therefore a transcranial approach can achieve complete removal of the mucocèle with wide exposure, good cosmetic result, avoiding possible relapses (26). In cases of FGM complicated by subdural empyema, as in our case, among the possible extensive surgical strategies, craniotomy is the most effective surgical procedure, although the so called decompressive craniectomy is also advocated in some conditions to reduce the intracranial hypertension (38, 39).

Our experience highlights the use of an internal remodelling cranioplasty consisting of surgical removal of the expanded thinned out outer table of the frontal sinus (that must be obliterated) in order to restore the internal contour of the forehead, so allowing a new frontal swelling lobe adaptation to the new gained intracranial space.

We call frontal "internal decompressive craniectomy with craniotomy" a wide bicoronic bone flap,

with frontal sinus all-embracing complete drilling of the inner wall. Suction of the fluid collection in the subdural space, surgical removal of the debris, local irrigation with saline and wide spectrum antibiotics and, mostly, decompression of the brain swelling, are effectively allowed by this approach.

In our case the complete removal of the posterior wall of the giant frontal sinuses allowed us to gain precious space for the surrounding brain, consisting of at least 1 cm or more frontally and producing the same "functional" biomechanical effect of the well known classic frontal "decompressive craniectomy", with consistent reduction of intracranial pressure and immediate neurological improvement.

Finally the "cranialization" with wide dural patch, muscle, gelfoam and fibrin glue of the bottom and the anterior wall of the sinus, allows anatomical and functional separation between the brain and the paranasal environment.

Differently from the decompressive craniectomy, which is a double stage procedure, the "functional decompressive craniectomy with craniotomy" is one stage surgery with all the related advantages: 1) less anaesthetics administration; 2) less overall duration of the operating procedure; 3) less bleeding or cortical brain injury secondary to manipulation of chronic local flogistic reorganization.

Conclusions

To the Authors knowledge this is the first case of GFM with SDE reported in the literature.

Concerning the GFM, since the primary goals of surgery are to eradicate the mass, with minimal morbidity and to prevent recurrence, several factors must be considered before deciding the surgical approach. The exposition of the mass must be wide, the cosmetic outcome must be satisfactory and the reconstruction of the region must be completed in one stage. During preoperative evaluation, CT scans and MR images are essential in assessing the presence and the nature of the above mentioned lesions. Because the orbit is the most frequently affected region, early drainage must be performed to avoid possible damage to the orbit with secondary visual impairments.

Despite improvements in antibiotic therapies, sinusitis still carries a risk of serious and potentially fatal complications. Aggressive search for complications and early intracranial imaging are the most important factors for the early diagnosis of SDE. The possibility of SDE or brain abscess should be considered when evaluating a patient with severe headache, rhinorrhea and fever, particularly if there is a history of recent sinus or otitic infection.

Although rare, the clinical course of SDE is fulminating and potentially life-threatening, unless treated promptly with surgery and appropriate intravenous antibiotics.

Moreover, as in our case, in patients with excessive intracranial involvement, in whom drainage is insufficient, the lesion might be removed through a wide craniofacial approach. The "internal decompressive craniectomy with craniotomy" with cranialization of the frontal sinuses is consistent with wide, stable and immediate decompression.

Cooperation among the otorhinolaryngologist, neurologist and neurosurgeon is critical in the pre-operative assessment.

Early recognition and treatment are essential to reduce subsequent morbidity and mortality.

Disclaimer section

The authors do not report any conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

REFERENCES

- Akiyama M, Iamamoto N, Hashigucci K. Frontal mucocele presenting as a subcutaneous tumour on the forehead. *Dermatology*. 1999;199:263-4.
- Hazan A, BlinP, Peytral C. *et al.* Apropos of 2 cases of mucocele with major ophthalmologic complications treated by endoscopic endonasal surgery. *Ann Otolaryngol Chir Cervicofac*. 1996;113:92-8.
- Serrano E, Pessey JJ, Lacomme Y. Sinus mucocoeles: diagnostic and surgical aspects (apropos of 8 cases treated with rhino-surgical endoscopy). *Acta Otorhinolaryngol Belg*. 1992;46:287-92.
- Diaz F, Latchow R, Duvall AJ 3rd, Quick CA, Erickson DL. Mucocoeles with intracranial and extracranial extensions. Report of two cases. *J Neurosurg*. 1978;48:284-8.
- Stankiewicz JA. Sphenoid sinus mucocele. *Arch Otolaryngol Head Neck Surg*. 1989;115:735-40.
- Spencer WR. Imaging quiz case 1. *Arch Otolaryngol Head Neck Surg*. 1998;124:1046-8.
- Steven CM, Jorge DL, Robert HM. Mucocoeles of the maxillary sinus. *Otolaryngol Head Neck Surg*. 1997;117:18-21.
- Chia-Hsiang Fu, Kai-Ping Chang, Ta-Jen Lee. The difference in anatomical and invasive characteristics between primary and secondary paranasal sinus mucocele. *Otolaryngology-Head and Neck Surgery*. 2007;136:621-625.
- Nakaiama T, Mori K, Maeda M. Giant pyocoele in the anterior cranial fossa. Case report. *Neurol Med Chir*. 1998;38:3499-502.
- Eufinger H. and Machtens E. Purulent pansinusitis, orbital cellulitis and rhinogenic intracranial complications. *J Craniomaxillofac Surg*. 2001;29:111-117.
- Weber R, Draf W, Keerl R, Constantinidis J. Aspect of frontal sinus surgery. III: Indications and results of osteoplastic frontal sinus operation. *HNO*. 1995;43:414-420.
- Osborn MK, Steinberg JP. Subdural empyema and other suppurative complications of paranasal sinusitis. *Lancet Infect Dis*. 2007;7:62-7.
- Pompili A, Mastrostefano R, Caroli F. *et al.* Mucocele of neurosurgical interest: clinical considerations on five cases. *Acta Neurochir (Wien)*. 1990;102:114-21.
- Sartor K, Karnaze MG, Winthrop ID. *et al.* MR imaging in infra-, para- and retrosellar mass lesions. *Neuroradiology*. 1987;29:19-29.
- Prandini MN, Tella OI, Lacanna SN, Antunes AC, Roithmann R. Giant mucocele: neurosurgical view. Report of two cases. *Arq Neuropsiquiatr*. 2005;63:535-8.
- Chiarini L, Nocini PF, Bedogni A, Consolo U, Giannetti L, Merli GA. Intracranial spread of a giant frontal mucocele: case report. *Br J Oral Maxillofac Surg*. 2000;38:637-640.
- Clayman GL, Adams GL and Paugh DL. Intracranial complications of paranasal sinusitis: a combined institutional review. *Laryngoscope*. 1991;101:234-239.
- Quraishi H, Zevallos JP. Subdural empyema as a complication of sinusitis in the pediatric population. *Int J Pediatr Otorhinolaryngol*. 2006;70:1581-1586.
- Singh B, Van Dellen J, Ramjetten S, Maharaj T. Sinogenic intracranial complications. *J Laryngol Otol*. 1995;109:945-950.
- Lang E, Curran A, Patil N, Walsh R, Rawluk D, Walsh M. Intracranial complications of acute frontal sinusitis. *Clin Otolaryngol*. 2001;26:452-457.
- Migirov L, Duvdevani S, Kronenberg J. Otogenic intracranial complications: a review of 28 cases. *Acta Otolaryngol*. 2005;125:819-822.
- Aoki N, Sakai T, Oikawa A, Takizawa T, Shishido T. Infected subdural effusion associated with resolving subdural hematoma. *Neurol Med Chir*. 1997;37: 637-9.
- Ong YK, Tan HK. Suppurative intracranial complications of sinusitis in children. *Int J Pediatr Otorhinolaryngol*. 2002;66:49-54.
- Rich PM, Deasy NP, Jarosz JM. Intracranial dural empyema. *Br J Radiol*. 2000;73:1329-1336.
- Conlon BJ, Curran A and Timon CV. Pitfalls in the determination of intracranial spread of complicated suppurative sinusitis. *J Laryngol Otol*. 1996;110:673-675.
- Baker AS. Role of anaerobic bacteria in sinusitis and its complications. *Ann Otol Rhinol Laryngol*. 1991; Suppl 154:17-22.
- Jones NS, Walker JL, Bassi S, Jones T, Punt J. The intracranial complications of rhinosinusitis: can they be prevented? *Laryngoscope*. 2002;112:59-63.
- Verbon A, Husni RN, Gordon SM. *et al.* Pott's puffy tumor due to *Haemophilus influenzae*: case report and review. *Clin Infect Dis*. 1996;23:1305-1307.

29. Brändle N, Zehnder M, Weiger R, Waltimo T. Impact of growth conditions on susceptibility of five microbial species to alkaline stress. *J Endod.* 2008;34:579-82.
30. Brook I. *Prevotella intermedia* meningitis associated with cerebrospinal fluid leakage in an adolescent. *Pediatr Infect Dis J.* 2003;22:751-3.
31. Duncan MJ. Oral microbiology and genomics. *Review. Periodontol.* 2000 (2005);38:63-71.
32. Fritschi BZ, Albert-Kiszely A, Persson GR. *Staphylococcus aureus* and other bacteria in untreated periodontitis. *J Dent Res.* 2008;87:589-93.
33. Kai A, Cooke F, Antoun N, Siddharthan C, Sule O. A rare presentation of ventriculitis and brain abscess caused by *Fusobacterium nucleatum*. *J Med Microbiol.* 2008;57:668-71.
34. Sassone LM, Fidel RA, Faveri M, Guerra R, Figueiredo L, Fidel SR, Feres M. A microbiological profile of symptomatic teeth with primary endodontic infections. *J Endod.* 2008;34:541-5.
35. Lefebvre L, Metellus P, Dufour H, Bruder N. Linezolid for treatment of subdural empyema due to *Streptococcus*: case reports. *Surg Neurol.* 2008;8.
36. Koike Y, Tokoro K, Chiba Y, Suzuki SI, Murai M, Ito H. Intracranial extension of paranasal sinus mucocele: two case reports. *Surg Neurol.* 1996;45:44-8.
37. Delfini R, Missori P, Iannetti G, Ciapetta P, Cantore P. Mucocoeles of the paranasal sinuses with intracranial and intraorbital extension: report of 28 cases. *Neurosurgery.* 1993;32:901-906.
38. Nathoo N, Nadvi SS, Gouws E, van Dellen JR. Craniotomy improves outcomes for cranial subdural empyemas: computed tomography-era experience with 699 patients. *Neurosurgery.* 2001;49:872-877.
39. Ong YK, Goh KY, Chan C. Bifrontal decompressive craniectomy for acute subdural empyema. *Childs Nerv Syst.* 2002;18:340-343.

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