Endoscopic-assisted laser therapy for extensive rhinoscleroma

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Background
Scleroma is a granuloma affecting mainly the nose, and may extend to affect the pharynx, the larynx, the lacrimal apparatus, and many soft tissue structures surrounding the nose. In Egypt, as in many other Eastern countries, scleroma is the most common nasal granuloma diagnosed clinically and pathologically. Although it mainly affects the anterior part of the nose, it is not uncommon to find cases with extensive lesions that extend to occupy the entire nasal cavity, and also to the sinuses and the orbit. Active rhinoscleroma is usually treated medically, and surgical intervention is carried out in the fibrotic stage.

Theory
The aim of this study was to determine the role of CO_2 laser in the management of extensive active rhinoscleroma.

Materials and methods
This is a prospective study that included 24 patients with extensive rhinoscleroma in the active phase, that is, patients had almost complete nasal obstruction with mouth breathing. Patients were divided into two groups: group A (12 cases), who were managed only with medical treatment, and group B (12 cases), who were managed with CO_2 laser debulking, concurrently with medical treatment.

Results
Patients were followed up clinically for nasal blockage and histologically to assess the activity of the disease; group B showed earlier relief of nasal obstruction and needed less medical treatment to reach the inactive stage.

Conclusion
CO_2 laser debulking is a good option in treating extensive rhinoscleroma, even in the active stage, as it provides rapid relief of nasal blockage and decreases the time needed for medical treatment, reducing the possible side effects of antibiotics.

Keywords:
CO_2 laser, debulking, rhinoscleroma

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Introduction

Rhinoscleroma is a chronic progressive inflammatory disease of the upper respiratory tract [1]. The name rhinoscleroma was first used by Von Hebra and Kaposi in 1870, when describing a lesion in the nose that they labeled as a form of sarcoma [2]. In 1877, Mikulicz described the histological feature of this disease in detail and established its non-neoplastic inflammatory nature [3].

Von Frisch identified the causative agent of this lesion in 1882 as a Gram-negative coccobacillus, now known as *Klebsiella rhinoscleromoatis* [4].

Rhinoscleroma may present with mass lesions in the respiratory tract anywhere from the nose to the trachea [5]; the nasal cavity is most often affected (95–100%), but lesions may also develop in the larynx (15–40%), the nasopharynx (18–43%), the oral cavity, paranasal sinuses (28%), soft tissue of the lip and nose, trachea (12%), bronchi (2–7%), and rarely the orbit or the middle ear [6]. The condition in often indolent, progressive, prone to recurrence, and extremely difficult to cure; no racial predilection exists.

Rhinoscleroma is widely but unevenly distributed worldwide, often occurring in focal specific geographical areas. A large endemic area is present in Eastern Europe, extending into the Ukraine and around the black and caspian seas [7]; the disease has been reported in many countries in the middle east, tropical Africa, India, Southeast Asia, and Central and South America [8].

Rhinoscleroma is more common in developing countries and rural areas; there appear to be associations between poor hygiene, poor nutrition, and crowded living conditions and the development of rhinoscleroma.

Women are slightly more affected than men (about 1.3–1) and patients are commonly affected in the second and third decades of life [1].

Although the infectious agent has been characterized, the mechanism of infection and the pathophysiology of disease progression are poorly understood. Transmission has been proposed to occur through direct inhalation or inoculation by respiratory droplets, but only after prolonged contact [5].

Host factors may play a role in the development of the disease. Cellular immunity is impaired in the affected patients; studies in patients with rhinoscleroma have shown alterations in the ratio of CD4-positive lymphocytes (T-helper cells) to CD8-positive lymphocytes (cytotoxic T cell), with a marked increase in the latter type. Patients have also shown an impaired response of their CD4 lymphocytes to interleukin-2 and also a reduced proliferative response to concavalin A, a T-lymphocyte mitogen [9].

Classically, there are three clinical and histopathological stages of rhinoscleroma: catarrhal (atrophic), granulomatous, and sclerotic.

The histological findings are more characteristic and diagnostic in the proliferative stage; the catarrhal stage has no specific feature that the pathologist can identify. If clinically suspected, a nasal swab for culture to isolate the micro-organisms would confirm the diagnosis [10].

Other granulomatous, neoplastic, and infectious lesions must also be considered in the differential diagnosis. These include sarcoidosis, Wegener's granulomatosis, lethal midline granuloma, vasculitis, lymphoma, basal cell cancer, verrucous carcinoma, actinomycosis, paracoccidioidomycosis, leishmaniasis, leprosy, tuberculosis, sporotrichosis, syphilis, rhino-oporidiosis, nasopalatine duct cyst, and Rosai–Dorfman disease.

Establishment of the diagnosis is often challenging; when rhinoscleroma is suspected, a brush biopsy specimen of the nasal or the respiratory tract mass or an incisional biopsy of an easily accessible lesion should be sent for cytology and culture. A positive culture of *K. rhinoscleromatis* on blood or MacConkey agar is diagnostic of rhinoscleroma but only 50–60% of patients are culture-positive. The bacteria may also be identified using periodic acid-Schiff, Giemsa, Gram, or Warthin–Starry silver stains. The histology changes according to the stage of the disease but is characteristically marked by subepithelial Mikulicz cells and transformed plasma cells with Russell bodies. These pathologic findings, along with pseudoepitheliomatous hyperplasia, are usually present in the hypertrophic or the granulomatous stage [5].

Treatment of rhinoscleroma has been attempted for many decades using various antibiotics and chemotherapeutic agents without much success. Only a few of these drugs have been shown to be effective to a limited degree. Complete cure of cases of rhinoscleroma treated by the prolonged use of specific antibiotics is uncertain [11].

Currently, the recommended treatment of rhinoscleroma consists of a combination of surgical debridement and long-term antibiotic therapy. Ciprofloxacin, tetracycline, and topical and systemic refampicin and gemifloxacin have been used with success [12].

Most of the studies in the literature have suggested continuation of treatment for at least a month after clinical resolution has been documented [13].

There are several indications for surgery in rhinoscleroma, including removal of nasal granulations, bronchoscopic dilatation, bipolar coagulation of skin lesions, tracheostomy, and repair of pharyngeal stenosis [14].

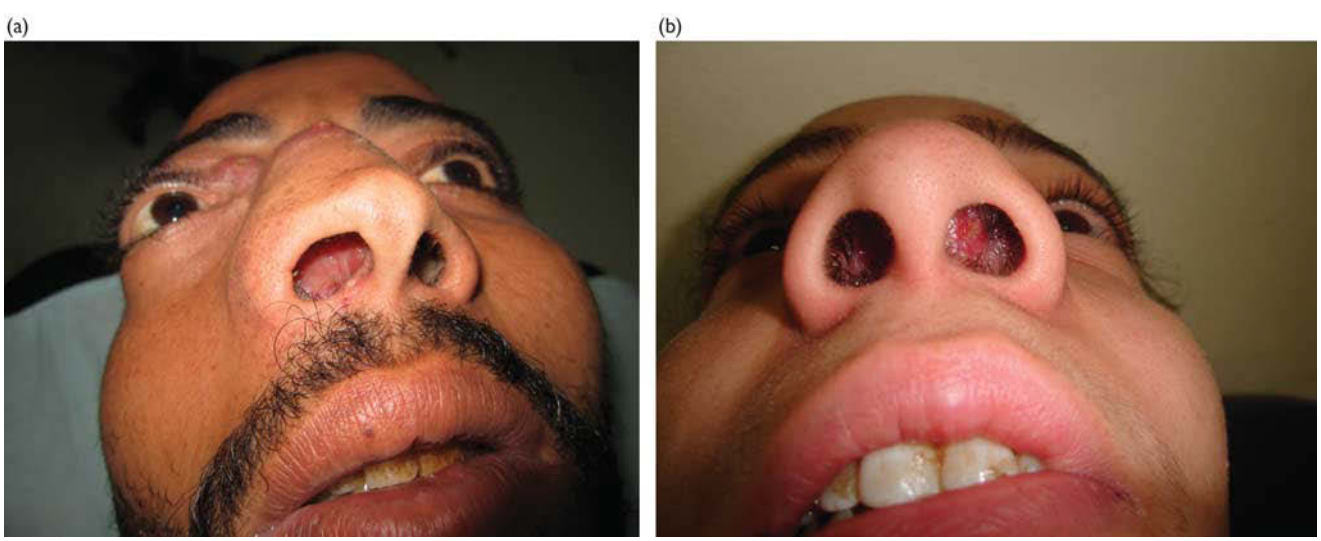
Materials and methods

This is a prospective study that included 24 patients with extensive rhinoscleroma in the active phase; the study was carried out in the period between March 2009 and October 2011, in the Department of Otorhinolaryngology, Faculty of Medicine, Cairo University (Cairo, Egypt).

The inclusion criteria were as follows:

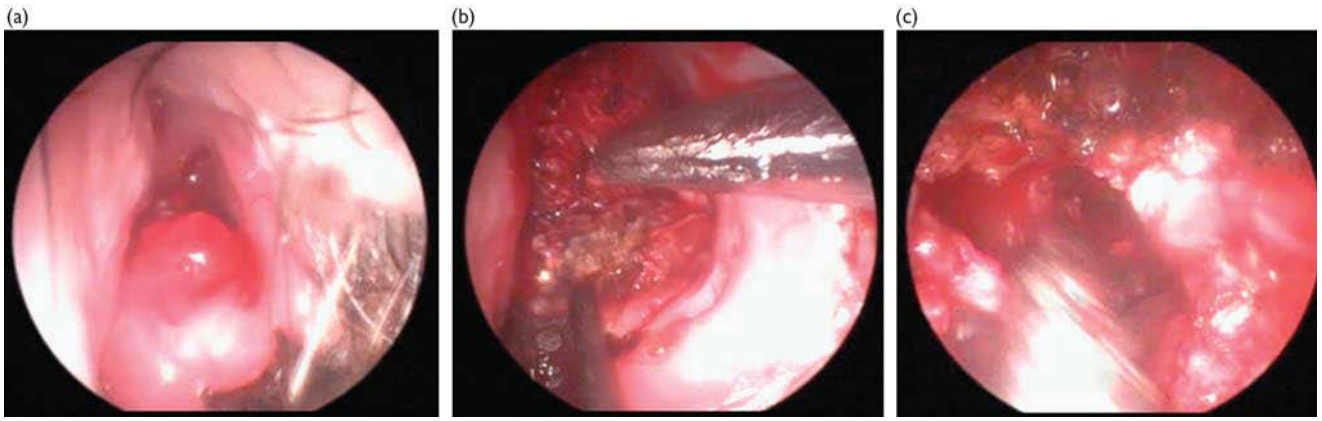
(1) age over 16 years.
(2) informed consent.
(3) active rhinoscleroma proved pathologically, and patients presented with severe bilateral nasal obstruction (Fig. 1), with no other cause.

Figure 1



(a and b) Rhinoscleroma with a complete nasal block.

Figure 2



(a) Active nasal scleroma, (b) laser ablation procedure, (c) nose open to the choana.

(4) no previous treatment.
(5) absent sinus involvement (by CT).

All patients were subjected to detailed history taking, medical examination, computed tomography nose and paranasal sinuses coronal cuts, and nasal biopsy to confirm the diagnosis.

Patients were randomly divided into two groups. (a) Group A: patients in group A (12) were treated medically using rifampicin 300 mg twice daily for 3 months. (b) Group B: patients in group B (12) were treated both medically and surgically; medical treatment was the same as in group A, whereas surgical treatment involved endoscopic-assisted CO_2 laser debulking (power 10, continuous mode, super pulsed CO_2 laser); the procedure took 40 min on average, and not much bleeding was encountered (Fig. 2).

For both groups, the follow-up was for at least 6 months, which included the following:

(1) Questionnaire about nasal symptoms (mainly nasal blockage) every 1 month; patients were scored according to improvement in nasal obstruction as follows:

No improvement	0
Mild improvement	1
Moderate improvement	2
Total relief	3

(2) Nasal biopsy after 3 and 6 months to assess pathological cure and recurrence.

Note: patients were followed till they became clinically free with improved nasal obstruction, and pathologically free proved by negative nasal biopsy.

The 24 patients included in this study fulfilled the criteria of management and follow-up; those who failed to comply with the treatment or follow-up were excluded from the study.

Results

The study included 24 patients (15 women and nine men). Group A included eight women and four men and group B included seven women and five men, ranging in age between 16 and 42 years.

Table 1 Analog score 1 × group

	Group		
	Group A	Group B	Total
Analog score 1			
0			
Count	8	0	8
% within group	66.7	0.0	33.3
1			
Count	4	4	8
% within group	33.3	33.3	33.3
2			
Count	0	5	5
% within group	0.0	41.7	20.8
3			
Count	0	3	3
% within group	0.0	25.0	12.5
Total			
Count	12	12	24
% within group	100.0	100.0	100.0
χ^2 tests	Value	*d.f.*	*P* value
Pearson's χ^2	16.000	3	0.001
Likelihood ratio	22.181	3	0.000
Linear-by-linear association	14.049	1	0.000
Number of valid cases	24		

Eight cells (100.0%) have an expected count less than 5. The minimum expected count is 1.50.

The analog scores for each group, every month, are presented in the following tables (Tables 1–6).

The results of pathological examination are presented in the following tables (Tables 7 and 8).

Discussion

Despite being an old disease, some aspects of scleroma are still not well established; as medical treatment with different antimicrobial agents remains the cornerstone in any therapy, many types of antibiotics have been used, with different degrees of success. N'Gattia *et al.* [15] used streptomycin, thiobenicol, or ciprofloxacin, whereas Gaafar *et al.* [14] used a combination of trimethoprim–sulfamethoxazole 400 mg and

Table 2 Analog score 2 × group

	Group		
	Group A	Group B	Total
Analog score 2			
0			
Count	1	0	1
% within group	8.3	0.0	4.2
1			
Count	4	0	4
% within group	33.3	0.0	16.7
2			
Count	6	3	9
% within group	50.0	25.0	37.5
3			
Count	1	9	10
% within group	8.3	75.0	41.7
Total			
Count	12	12	24
% within group	100.0	100.0	100.0
χ^2 tests	Value	*d.f.*	*P* value
Pearson's χ^2	12.400	3	0.006
Likelihood ratio	15.312	3	0.002
Linear-by-linear association	10.837	1	0.001
Number of valid cases	24		

Six cells (75.0%) have an expected count less than 5. The minimum expected count is 0.50.

Table 3 Analog score 3 × group (crosstab)

	Group		
	Group A	Group B	Total
Analog score 3			
1			
Count	2	0	2
% within group	16.7	0.0	8.3
2			
Count	3	1	4
% within group	25.0	8.3	16.7
3			
Count	7	11	18
% within group	58.3	91.7	75.0
Total			
Count	12	12	24
% within group	100.0	100.0	100.0
χ^2 tests	Value	*d.f.*	*P* value
Pearson's χ^2	3.889	2	0.143
Likelihood ratio	4.715	2	0.095
Linear-by-linear association	3.696	1	0.055
Number of valid cases	24		

Four cells (66.7%) have an expected count less than 5. The minimum expected count is 1.00.

rifampicin 300 mg twice daily for 3 months; since 2003, this has been replaced by ciprofloxacin 500 mg twice daily for 3 months. Moraes *et al.* [12] used a combination of gemifloxacin and tetracyclins. In our study, rifampicin 600 mg daily for 3 months was used due to the availability and the low cost of the drug.

In many cases, surgery is required in addition to medical treatment to relieve airway obstruction in the nasal cavity, pharynx, larynx, or trachea [16].

Introduction of CO_2 laser allowed removal of adhesion with less blood loss and lower incidence of fibrous tissue formation; usually, a CO_2 laser is used to relieve nasal obstruction in the fibrotic stage of rhinoscleroma [17].

Table 4 Analog score 4 × group (crosstab)

	Group		
	Group A	Group B	Total
Analog score 4			
1			
Count	0	1	1
% within group	0.0	8.3	4.2
2			
Count	4	3	7
% within group	33.3	25.0	29.2
3			
Count	8	8	16
% within group	66.7	66.7	66.7
Total			
Count	12	12	24
% within group	100.0	100.0	100.0
χ^2 tests	Value	*d.f.*	*P* value
Pearson's χ^2	1.143	2	0.565
Likelihood ratio	1.530	2	0.465
Linear-by-linear association	0.126	1	0.723
Number of valid cases	24		

Four cells (66.7%) have an expected count less than 5. The minimum expected count is 0.50.

Table 5 Analog score 5 × group (crosstab)

	Group		
	Group A	Group B	Total
Analog score 5			
1			
Count	1	2	3
% within group	8.3	16.7	12.5
2			
Count	5	2	7
% within group	41.7	16.7	29.2
3			
Count	6	8	14
% within group	50.0	66.7	58.3
Total			
Count	12	12	24
% within group	100.0	100.0	100.0
χ^2 tests	Value	*d.f.*	*P* value
Pearson's χ^2	1.905	2	0.386
Likelihood ratio	1.955	2	0.376
Linear-by-linear association	0.080	1	0.777
Number of valid cases	24		

In our study, we used the CO_2 laser in addition to medical treatment to relieve nasal obstruction, and attempted to decrease recurrence and hence the period of antimicrobial therapy.

It can be seen from Fig. 3 that group B showed a marked improvement in nasal obstruction in the early months compared with group A; this was maintained up to the fourth month, when the analog score of both groups became almost similar with slight advance in group B. This can be attributed to the relatively high incidence of recurrence (Fig. 4); pathology after the third month was positive in 30% of the patients in group A and 22% of the patients in group B. This increased at the end of the sixth month to 40% in group A and 30% in group B.

The recurrence rates are highly variable in the literatures, ranging from very rare [18] up to 25% [14]; the durations of follow-up needed to detect the recurrence also vary markedly. In our study, the pathological results after

Table 6 Analog score 6 × group (crosstab)

	Group		
	Group A	Group B	Total
Analog score 6			
1			
Count	3	3	6
% within group	25.0	25.0	25.0
2			
Count	2	1	3
% within group	16.7	8.3	12.5
3			
Count	7	8	15
% within group	58.3	66.7	62.5
Total			
Count	12	12	24
% within group	100.0	100.0	100.0

χ^2 tests	Value	*d.f.*	*P* value
Pearson's χ^2	0.400	2	0.819
Likelihood ratio	0.407	2	0.816
Linear-by-linear association	0.054	1	0.816
Number of valid cases	24		

Four cells (66.7%) have an expected count less than 5. The minimum expected count is 1.50.

Table 7 Path 3 × group (crosstab)

	Group		
	Group A	Group B	Total
Path 3			
Negative			
Count	9	10	19
% within group	75.0	83.3	79.2
Positive			
Count	3	2	5
% within group	25.0	16.7	20.8
Total			
Count	12	12	24
% within group	100.0	100.0	100.0

χ^2 tests	Value	*d.f.*	*P* value	*P* value	*P* value
Pearson's χ^2	0.253	1	0.615		
Continuity correction	0.000	1	1.000		
Likelihood ratio	0.254	1	0.614		
Fisher's exact test				>0.999	0.500
Linear-by-linear association	0.242	1	0.623		
Number of valid cases	24				

Computed only for a 2 × 2 table.
Two cells (50.0%) have an expected count less than 5. The minimum expected count is 2.50.

6 months indicated that scleroma had a higher rate of recurrence than expected. It is difficult to compare the results in different countries as rhinoscleroma is associated with many epidemiologic and immunologic factors that contribute to endemicity and severity.

The statistical paragraph in materials and methods

Data were statistically described in terms of mean ± SD, median and range, or frequencies (number of cases) and percentages when appropriate. Comparison of numerical variables between the study groups was carried out using the Mann–Whitney *U*-test for independent samples. For comparison of categorical data, the χ^2-test was used. The exact test was used when the expected frequency was less than 5. *P* values less than 0.05 were considered

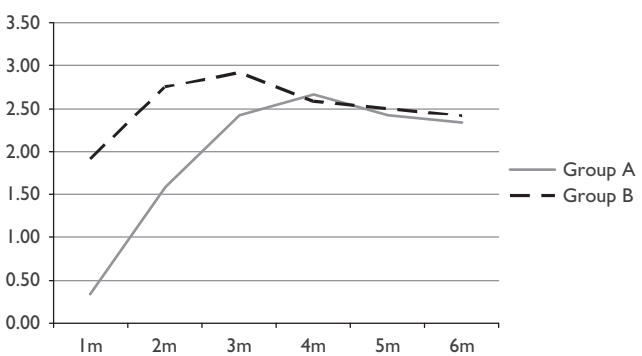
Table 8 Path 6 × group (crosstab)

	Group		
	Group A	Group B	Total
Path 6			
Negative			
Count	8	9	17
% within group	66.7	75.0	70.8
Positive			
Count	4	3	7
% within group	33.3	25.0	29.2
Total			
Count	12	12	24
% within group	100.0	100.0	100.0

χ^2 tests	Value	*d.f.*	*P* value	*P* value	*P* value
Pearson's χ^2	0.202	1	0.653		
Continuity correction	0.000	1	1.000		
Likelihood ratio	0.202	1	0.653		
Fisher's exact test				>0.999	0.500
Linear-by-linear association	0.193	1	0.660		
Number of valid cases	24				

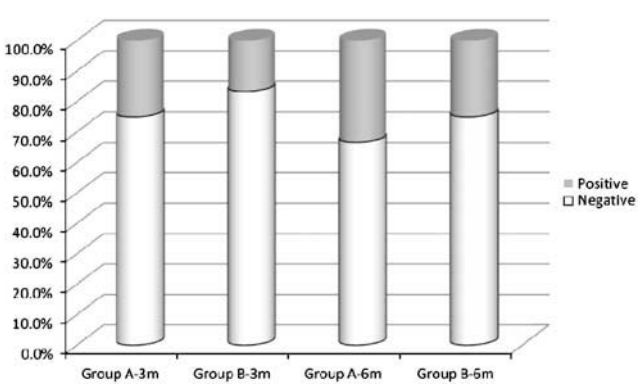
Computed only for a 2 × 2 table.
Two cells (50.0%) have an expected count less than 5. The minimum expected count is 3.50.

Figure 3



Mean analog scores of the two study groups over the study period.

Figure 4



Results of pathological examination of the two study groups over the study period.

statistically significant. All statistical calculations were carried out using computer programs statistical package for the social science (SPSS; SPSS Inc., Chicago, Illinois, USA) version 15 for Microsoft Windows.

Conclusion

(1) Endoscopic-assisted CO_2 laser in combination with medical treatment is a good option for the treatment of patients with active rhinoscleroma with severe bilateral nasal obstruction, as it provided better and early relieving of the nasal block; it is also associated with a lower recurrence rate and thus a smaller duration of antimicrobial treatment.
(2) Scleroma is a highly recurrent disease that needs a long follow-up duration, with frequent biopsies to exclude recurrence.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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