Primary tracheoscleroma

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SCLEROMA is a chronic granulomatous condition usually affecting upper part of the respiratory tract. It starts mostly in the nose and spreads to the naso-pharynx, throat, larynx, and trachea. Quevedo (1949) reviewing a series of 108 cases of scleroma from Guatemala observed that primary scleroma of larynx or trachea is very rare. Sinha and Pandhi (1969) reviewed 29 cases of scleroma and pointed out that the involvement of larynx and trachea was always secondary to nasal lesions. Handousa and Elwi (1958) from Cairo who saw about forty cases of scleroma per year, deny having seen a single case of isolated primary scleromatous lesion of trachea. Lund (1960) from Oxford, reported a fatal case of scleroma of trachea and observed that it was probably secondary to nasal and laryngeal lesions. This prompted us to report a case of isolated primary scleroma of trachea—a very rare entity.

Case records

B.R., 35 years, male, a rural farmer, was admitted to male medical ward of P.B.M. Hospital, Bikaner, on 10 April 1971 with the complaints of dysphagia and hoarseness of voice of 1½ years' duration. Dysphagia was both with solids and liquids and was painful and of severe degree for the past two weeks. Hoarseness was of insidious onset and non-progressive, He also suffered from cough with expectoration for the past one year, expectoration was mucopurulent, moderate in amount, not foul smelling, and was not associated with any crusts or haemoptysis.

The patient developed respiratory distress and noisy breathing two weeks before admission. He was diagnosed as a case of multiple cranial nerve palsy and was treated with oxytetracycline, orally injection of aminophylline and haematinics but obtained no relief. Tracheostomy was done at the bedside on 16 April 1971 to relieve the respiratory distress. He was given liquid diet. The patient was decannulated on 28 April 1971, and at the time of discharge from the hospital on 8 May 1971 he did not suffer from dysphagia or respiratory distress. The patient was readmitted on 21 May 1971 in male medical wards with the same complaints of hoarseness, dysphagia, cough with expectoration, and respiratory distress. There was no history of recurrent colds, fever, broncho-spasms, tuberculosis or syphilis in the past or in the family history of the patient. His socio-economic status was poor.

The examination of the patient revealed a middle-aged man of average build, and poor nourishment with poor hygiene. Mild anaemia present, afebrile. Respiratory rate 28 per minute. Inspiratory stridor present. There was no significant lymph node enlargement. Chest: normal breath sounds heard. Anterior and posterior rhinoscopy, examination of paranasal sinuses and throat was normal. Indirect laryngoscopic examination showed left vocal cord in the

https://doi.org/10.1017/S0022215100075836 Published online by Cambridge University Press

paramedian position with slight bowing on phonation. The mucosa over the first few tracheal rings was congested; no obstructive pathology could be seen.

Investigations

Blood: Haemoglobin 10 gm. per cent.
T.L.C., 7,200/cu.mm. D.L.C., Polymorphs 60. Lymphocytes 38, and Eosinophils 2 per cent.
E.S.R., 58 mm./1st hour westergren.
Kahn test, negative.

Urine and stool examination were normal.

C.S.F. examination: Proteins, 20 mg per cent. Sugar, 60 mg per cent. Chloride, 760 mg per cent. Microscopic examination showed 2–3 cells/high power field.

Skiagram of chest, normal. Barium swallow examination of oesophagus, normal, no pathology could be detected.

Laryngogram.

The structures of the larynx, i.e. pyriform fossae, ventricles, false and the true vocal cords and subglottic regions were normally outlined (Fig. 1).

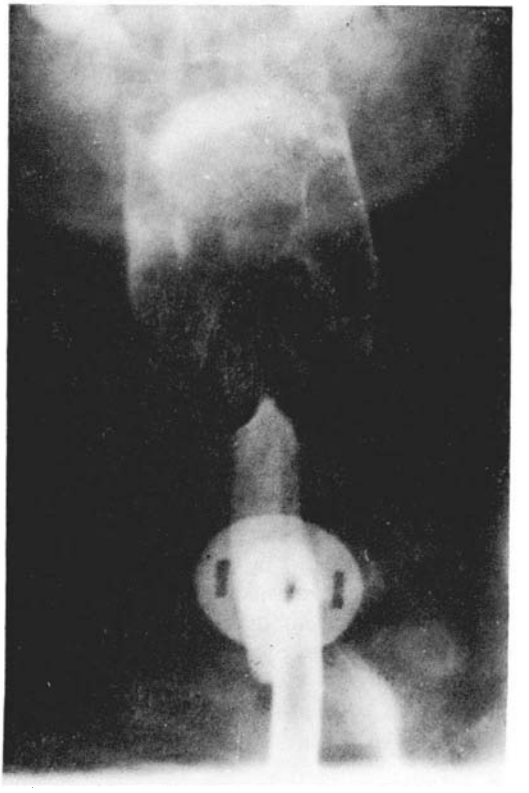


FIG. 1.
Laryngogram shows normal filling of pyriform fossae, ventricles, the false and true vocal cords are clearly outlined. Tracheostomy tube is in position.

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Tracheogram—outlines the stenosed part of the trachea and site of tracheostomy opening (Fig. 2).

Histopathology showed squamous metaplasia of the epithelium and collection of chronic inflammatory cells, viz. plasma cells, lymphocytes, monocytes, islands of Mickulicz cells with scanty cytoplasm and a few Russel bodies were seen. The histological picture was diagnostic of scleroma (Figs. 3, 4, and 5).

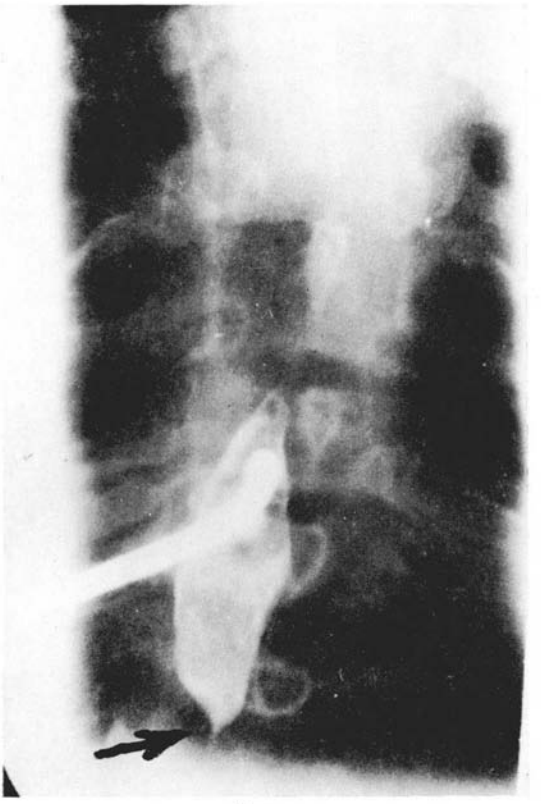


FIG. 2.

Tracheogram. The arrow shows the stenosis of the trachea caused by the scleromatous lesion. Nozzle of the syringe is in the tracheostoma to instil the radio-opaque medium.

Tissue and swab (from the lesion) for culture and sensitivity. Klebsiella rhinoscleromatis could not be isolated. Non-haemolytic streptococci sensitive to chloramphenicol and penicillin were isolated.

Tracheostomy at the level of third and fourth tracheal rings was undertaken under local anaesthesia. Jackson's tracheostomy tube of 10 mm. size could not be pushed down the trachea. A 7 mm. size Jackson's bronchoscope was passed through the tracheostoma and a circumferential fleshy mass was noticed to obstruct the lumen of the trachea at the level of 8th and 9th cartilage rings. The attempts at taking a biopsy from the lesion revealed rubbery hard consistency.

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The biopsy from the lesion was taken for histopathological examination. The subglottic region was also examined with the same bronchoscope inserted through the tracheostoma and no pathological lesion could be detected in the subglottic region and the segment of trachea above the tracheostoma. Fuller's tracheostomy tube, 24 size was inserted. Oesophagoscopy examination did not reveal any pathology in the oesophagus. Repeated dilatations were done at weekly

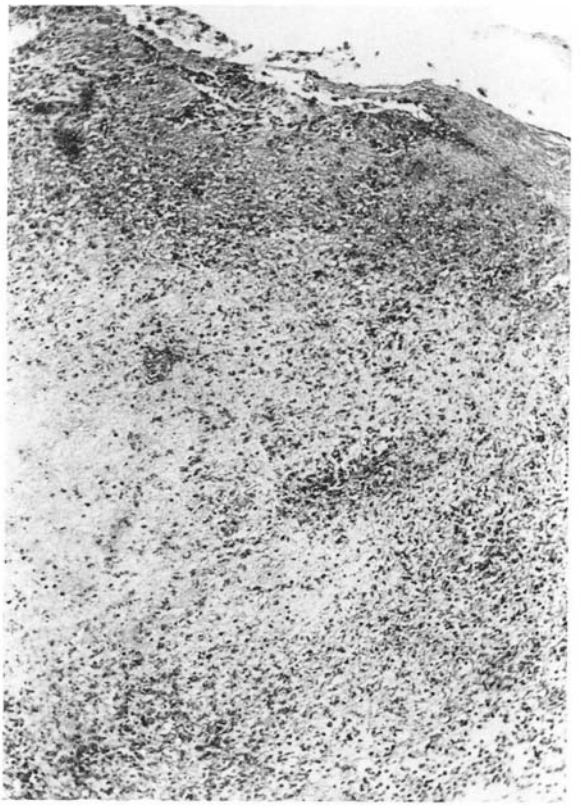


FIG. 3.

Histopathological section presents squamous metaplasia and chronic inflammatory cells, viz. plasma cells, lymphocytes, monocytes and islands of Mikulicz cells. (H.&E. 10 × 10.)

intervals with Jackson's bougies. Injections streptomycin 1 gm. and penicillin 4 lacs—I.U. were given daily intramuscularly along with haemantinics and aminophylline parentrally for three weeks. However, after the diagnosis of scleroma was established by the histopathological examination on 1 July 1971, the specific treatment with injection streptomycin 1 g. daily, aminophylline parenterally and haematinics was continued. The patient showed steady progress. However, on 8 August, 1971, the patient developed sudden respiratory distress and cyanosis. Local examination revealed the scleromatous lesion obstructing the lumen beyond the end of tracheostomy tube. An endotracheal tube of size 2

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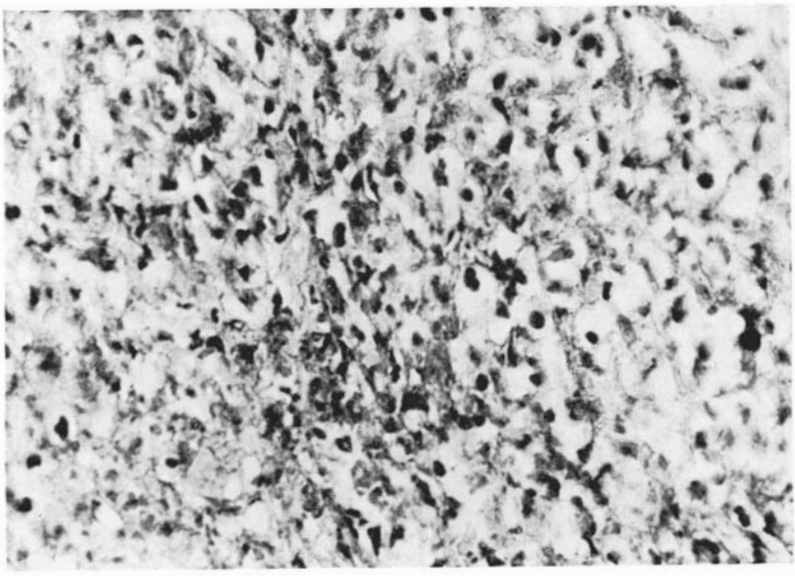


FIG. 4.
Section shows sheets of Mikulicz cells (M) with prominent central nucleus and scanty cytoplasm; a few Russel bodies (R) are seen. (H.&E. 45 × 10.)

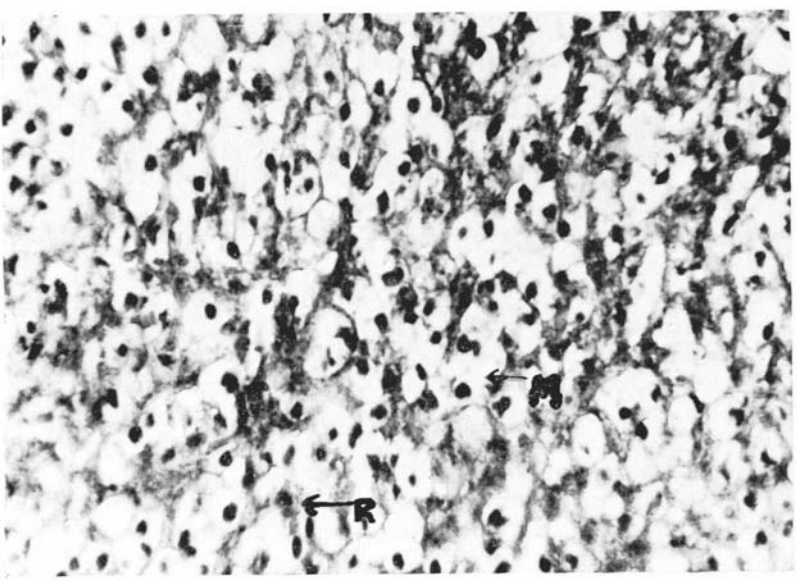


FIG. 5.
Section presents sheets of Mikulicz cells and chronic inflammatory cells. (P.A.S. 45 × 10.)

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was passed beyond the tracheal obstruction, through the tracheostoma (Fig. 6). Mucopurulent secretions were sucked from the bronchi with the help of the bronchoscope and oxygen inhalation was given. The size of endotracheal tube was gradually increased to number seven. The patient made satisfactory progress till the time of reporting under the regime of treatment mentioned above.

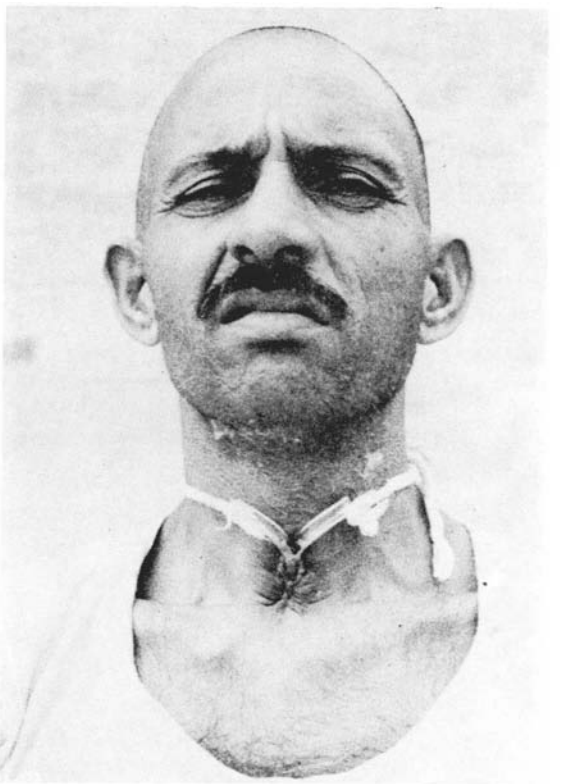


FIG. 6.
Photograph of the patient with endotracheal tube inserted through the tracheostoma.

Comments

Primary scleroma of the trachea is an extremely rare condition. The review of the available literature reveals one report by Dobrazanski (1951) presented at the Fourth International Congress of Otolaryngology (quoted by Negus, 1955). He, however, stressed that scleroma must be considered in a chronic granulomatous condition of the trachea. A case of scleroma has never been seen in Bikaner district by one of the authors during his stay of eight years even when the epidemiological conditions here are not unfavourable, viz. hot dry climate, poor hygiene, nutrition, and socio-economic status of the people residing in rural areas. The case under study hailed from the endemic areas of the state. However, the rubbery hard consistency of the lesion with scanty bleeding at the time of biopsy gave the suspicion of scleroma.

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The causative organisms could not be isolated in this case, probably because the patient had already received 23 gm. of streptomycin before the tissue and swab were innoculated. However, the histopathological studies were diagnostic of scleroma. The use of laryngeal dilators has been advocated in the treatment of laryngeal or tracheal lesions by Quevedo (1949) and Kakar *et al.* (1964-65). However, the use of endotracheal tube which could provide continued dilating effect and give satisfactory results if continued over a period of six to eight weeks are suggested by the authors for trial in such cases. It could also help epithelialization of the lesion under the umbrella of antibiotic therapy.

Summary

A case of primary scleroma of trachea has been reported. The patient responded satisfactorily with streptomycin and endotracheal intubation through the tracheostoma. The use of endotracheal tube to achieve a continuous dilatation of the lesion is suggested.

Acknowledgements

We are grateful to Dr. K. D. Gupta, M.D., Principal and Controller, S.P. Medical College and Associated Group of Hospitals, Bikaner, for permitting us to publish the hospital records. The authors are thankful to Dr. I. N. Ramdeo, M.D., Reader in Pathology for the histopathological studies of the case.

REFERENCES

DOBRZANSKI, A. (1951) *Proceedings of the 4th International Congress of Otolaryngology* (1949), vol. 1, p. 393, British Medical Association, London. Quoted by V. E. Negus (1955).

HANDOUSA, A., and ELWI, A. M. (1958) *Journal of Laryngology and Otology*, **72**, 32.

KAKAR, P., SINHA, A., and SANGWAN, R. S. (1964-5) *Indian Journal of Otology*, **16**, 164.

LUND, W. S. (1960) *Journal of Laryngology and Otology*, **74**, 791.

PANDHI, S. C. (1966) *Aetiopathogenesis of scleroma*. Master's thesis, A.I.I.M.S., New Delhi.

QUEVEDO, J. (1949) *Annals of Otology, Rhinology and Laryngology*, **58**, 613.

SINHA, A., and PANDHI, S. C. (1969) *Eye, Ear, Nose and Throat Monthly*, **48**, 109.

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https://doi.org/10.1017/S0022215100075836 Published online by Cambridge University Press

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