

View from Within: Radiology in Focus

Non-occupational recurrent bilateral pneumoparotitis in an adolescent

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Abstract

This is a case report of recurrent bilateral parotid swelling with intraglandular gaseous bubbles in a 14-year-old boy, together with a review of the literature on this condition, which is usually called pneumoparotitis. The disorder has been reported as an occupational hazard in wind instrument players and glass-blowers and also as a rare non-occupational disease, mainly in adolescents and often associated with psychological problems.

Introduction

Recurrent parotitis due to pneumatic inflation of the parotid glands has been reported as an occupational hazard in glass-blowers (Reitlinger, 1964; Rysenaer *et al.*, 1964), wind instrument players (Rysenaer *et al.*, 1964) and balloon-blowers (Rupp, 1963), as well as in a diver and a watch-keeper working under high barometric pressure (Watt, 1977). As a non-occupational disorder, it has been described (Hemphill, 1973) in a group of

French Foreign Legionaries who simulated mumps by blowing hard into a bottle, thus forcing air into the parotids and causing pneumoparotitis. It has also been reported in children playing with toy instruments (Tkacz, 1958), in a case of cough suppression (Aristy, 1982), in a hay-fever patient after forceful nose-blowing (Garber, 1987) and in a patient with buccinator hypotonia (Telfer and Irvine, 1989).

In addition to such sporadic cases, there are various reports of

TABLE I
 PNEUMOPAROTITIS IN PATIENTS WITH PSYCHOSOCIAL PROBLEMS

| Author(s) | Age (years) | Sex | Clinical evidence | Psychological background | Sialogram |
|--|-------------|-----|--|---|--|
| 1. Rupp (1963) | 11 | M | Recurrent bilateral pre-auricular swelling. Crepitation | Conflict with parents | Subcutaneous air, duct distortion |
| 2. Rysenaer <i>et al.</i> (1964) | 13 | F | Recurrent unilateral pre-auricular swelling subcutaneous emphysema | Nervous tic | Not stated |
| 3. Rysenaer <i>et al.</i> (1964) | Teens | F | Recurrent bilateral pre-auricular swelling | Nervous tic. Psychiatric treatment | Normal |
| 4. Greisen (1968) | 9 | M | Recurrent bilateral pre-auricular swelling. | Nervous tic | Subcutaneous air |
| 5. Calcaterra and Lowe (1973) | 14 | M | Recurrent unilateral pre-auricular swelling. Crepitation. | Adolescent adjustment reaction | Subcutaneous air, contrast extravasation |
| 6. Watt (1977) | - | M | Unilateral pre-auricular swelling. Subcutaneous emphysema | Abnormal behaviour, psychiatric treatment | Dilated Stensen's duct |
| 7. Sanchez <i>et al.</i> (1980) | 15 | F | Recurrent bilateral pre-auricular swelling | Self-instrumentation. Severe psychological problems | Normal |
| 8. Hadas (1982) | 16 | M | Pre-auricular tenderness | Adolescent adjustment reaction | Normal |
| 9. Markowitz-Spence <i>et al.</i> (1987) | 12 | F | Recurrent bilateral pre-auricular swelling | Adolescent adjustment reaction | Air in parotid glands and subcutaneous |
| 10. David and Kanga (1988) | 6.5 | F | Crepitant swelling over left pre-auricular area, cystic fibrosis | Nervous tic | Cystic dilatation with gaseous translucent areas |
| 11. Ferlito <i>et al.</i> | 14 | M | Recurrent bilateral pre-auricular swelling | Nervous tic | Air in the parotid glands |

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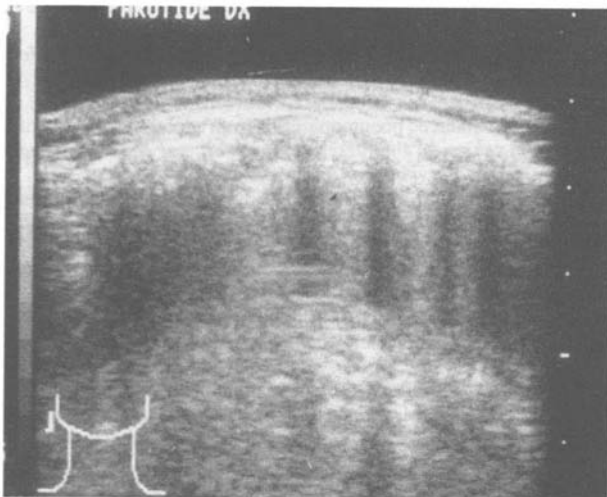


Fig. 1

Hyperechoic points with acoustic shadows for presence of air within the right parotid gland.

non-occupational pneumoparotitis associated by patient's age (almost all under 17 years) and abnormal psychological behaviour (Calcaterra and Lowe, 1963; Rupp, 1963; Rysenaer *et al.*, 1964; Greisen, 1968; Watt, 1977; Sanchez *et al.*, 1980; Hadas, 1982; Markowitz-Spence *et al.*, 1987; David and Kanga, 1988) (Table I). A similar such case is presented here, together with a review of the 10 cases previously reported in the literature.

Case report

A 14-year-old white boy presented in October 1989 with a history of recurrent bilateral parotitis. Relevant physiological features of the boy's background included thumb-sucking up to the age of 10 and a strange tongue sucking during sleep. The first episode had occurred at the age of 12 with swelling, pain and fever lasting one week. Months later, several similar episodes occurred. Oral examination revealed normal Stensen's ducts with fluid parotid saliva. Laboratory tests were consistent with inflammatory parotid swelling (amylasaemia = 1178 IU/L) without infection. During remission, ultrasound revealed normal parotid glands in size and morphology, with some hypo-echoic nodules (probably lymph nodes) in the glands and upper neck. Sialography revealed no abnormalities on either plain X-rays or after injection of contrast medium. Repeat laboratory tests revealed slight eosinophilia and higher than normal amylasaemia (555 IU/L). Tests for *Toxoplasma gondii*, Epstein Barr virus, paramyxovirus, cytomegalovirus, German measles and *Herpes simplex virus* antibodies were all negative. Fine needle biopsy of the parotid area revealed no abnormalities. Orthopantomography excluded odontogenic disorders.

The boy had been treated with antibiotics, anti-inflammatory drugs and corticosteroids with no improvement in the clinical course of recurrences.

He came under our observation during a relapse, when physical examination revealed bilateral painful swelling and oral examination showed normal Stensen's ducts with fluid saliva and no sign of Stensen's duct papilla stenosis. Ultrasound showed enlarged lymph nodes in the right upper neck, an enlarged right parotid gland with various hyperechoic areas and acoustic shadows due to the presence of gas (Fig. 1). Plain X-rays confirmed the presence of gas bubbles within the right parotid gland (Fig. 2), though it was impossible to establish whether they were in the duct system or parenchyma. After a review of the literature, the patient was re-evaluated and this revealed a nervous tic consisting of his puffing out his cheeks with closed lips.

Discussion

Under physiological conditions, air and oral secretions are

prevented from flowing back into the parotid duct and gland by various anatomical mechanisms:

- the duct orifice is slit-shaped and is sealed off by folds of mucosa when intra-oral pressure increases;
- buccinator muscle contraction closes the duct;
- cheek inflation increases duct angle both between the oral submucosa and buccinator muscle and between the latter and the subcutaneous tissue, alongside the masseter muscle.

When intra-oral pressure increases from 2–3 mmHg as in normal breathing, to 140–150 mmHg as in glass-blowing and wind instrument playing, air may be insufflated into the parotid glands, thus causing pneumoparotitis (Reitlinger, 1964; Rysenaer *et al.*, 1964). The condition seems prevalent in novice instrumentalists who blow into their instruments with 'full cheeks', but is seldom seen among those who blow with 'contracted cheeks' (Calcaterra and Lowe, 1963; Greisen, 1968).

A similar inflated-cheek increase in oral pressure probably lies behind the cases of non-occupational pneumoparotitis (Table I), which seem correlated by two aspects, *i.e.* the patient's age (10 cases under 17 years old, one not stated) and their psychological behaviour (ranging from nervous tics to adolescent adjustment reactions and even severe self-induced injury). These patients either voluntarily or unintentionally forced air into the parotid duct and glands, thus causing parotitis, often with superimposed bacterial infection.

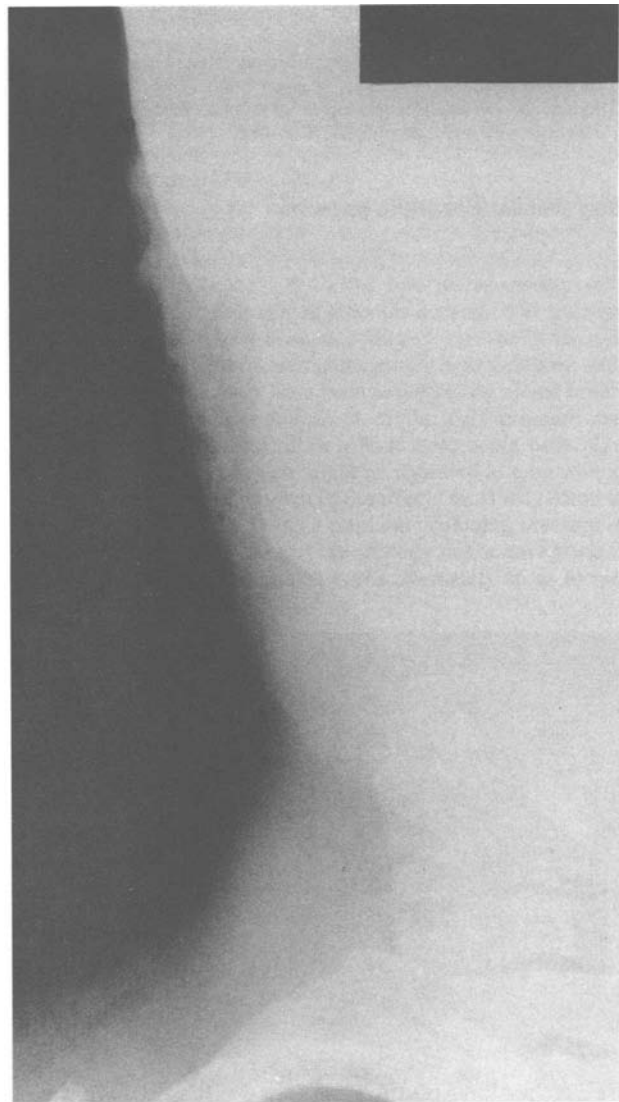


Fig. 2

Plain film of the right parotid gland shows many small transparent areas due to presence of air.

Clinical examination was helpful in diagnosing cases presenting crepitation for subcutaneous emphysema in the pre-auricular region, eyelid, mediastinum and upper thorax. In other cases, the clinician faced more underhand situations and reached the diagnosis only after discovering the patient's nervous tic or by means of instrumental investigations.

This pathogenetic mechanism has been confirmed by Calcaterra and Lowe (1963) on a patient invited to inflate his cheek with a water-soluble contrast medium in his mouth—fluoroscopy showed this material entering the parotid duct together with air bubbles.

The therapeutic implications of pneumoparotitis are obvious:— symptomatic treatment of recurrences (anti-inflammatory drugs and/or antibiotics);— acquisition of a correct blowing technique or suspension of activity in occupational forms; counsel on how to maintain normal intra-oral pressure and psychological support in non-occupational cases.

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