

Pneumoparotid: a rare cause of parotid swelling in a 7-year-old child

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Abstract. *Pneumoparotid: a rare cause of parotid swelling in a 7-year-old child. Problems/objectives:* Pneumoparotid is a rare condition in which air is insufflated through Stensen's duct into the parotid gland. This causes mostly painless swelling in the parotid region.

Case report: We report on a 7-year old girl presenting with painless unilateral swelling of the left parotid region, present for one month. Computer tomography showed the presence of air in the left parotid gland and in both the left and right Stensen's duct. A thorough history revealed the girl was insufflating the parotid glands intentionally as a response to stress. The patient was comforted and instructed to avoid insufflation manoeuvres, which resulted in complete resolution.

Conclusion: Pneumoparotid should be considered part of a differential diagnosis in children and adolescents presenting with painless uni- or bilateral swelling of the parotid gland. Supportive and conservative treatment is advocated.

Introduction

Swelling of the parotid region can be caused by both acute and chronic disorders. An overview of the differential diagnosis for parotid swelling in children is given in Table 1. The diagnosis of parotid enlargement in children should be based on an adequate history, physical examination, laboratory findings, imaging, and needle aspiration cytology when indicated. A thorough history could reveal exposure to infectious diseases and a positive family history of inflammatory disorders. Blood should be examined to determine blood counts, amylase, acute phase reactants, and immunological parameters like antinuclear- and anticytoplasmatic antibodies and rheumatoid factor.^{1,2} A tuberculin sensitivity test should be performed when indi-

cated. Imaging can consist of ultrasound, computer tomography, or MRI.¹ Pneumoparotid is the presence of air in the parotid gland, which may cause swelling of the gland and can be accompanied by inflammation. Pneumoparotid is a rare condition in children. We describe a self-induced pneumoparotid in a paediatric patient.

Case report

A 7-year old girl presented with left sided swelling of the parotid gland for one month. The swelling was not painful and the girl had not suffered from fever or other systemic symptoms. The swelling was continuously present and not influenced by food intake. Palpation of the gland showed a rather hard swelling on the left side and a mild swelling on the

right side. There was no crepitus. Intraoral examination showed purulent secretions from Stensen's duct when the parotid region was massaged. Microbiological examination of the secretions showed limited growth of *H. influenzae*. A blood sample revealed normal values for whole blood count, acute phase reactants, antinuclear- and anticytoplasmatic antibodies, and rheumatoid factor. The tuberculin sensitivity test was negative. Computer tomography showed bilateral dilated intraglandular and extraglandular ducts. A large amount of air was present in the left parotid gland and a small amount of air in the right parotid gland and duct (Figure 1). The diagnosis of pneumoparotid was made.

On careful questioning the girl admitted she had a habit of blowing out her cheeks, especially in

Table 1
Aetiology of swelling in the parotid region in children

Aetiology	Examples	Remarks
Infectious or Inflammatory ^{1,3}	Mumps Nontuberculous or tuberculous mycobacteria Cytomegalovirus Epstein-Barr virus HIV Influenza A Parainfluenza 1 and 3 Coxsackie virus Cat scratch disease Juvenile recurrent parotitis Obstructive sialoadenitis Sjögren's disease	Most common aetiology Rare condition in children Recurrent parotitis or parotid gland enlargement more common in children Wegener's disease
Systemic disorders ^{2,4}	Diabetes Hypothyroidism Liver failure Cushing's disease Drugs Allergy Bulimia nervosa	
First branchial cleft anomalies ^{2,5}	Infection of cyst or sinus in preauricular region or upper neck	
Vascular anomalies ^{1,2,5}	Haemangioma Lymphangioma	Most common vascular anomaly
Benign parotid tumour Malignant parotid tumours ^{1,3,6}	Pleomorphic adenoma Mucoepidermoid carcinoma Acinic cell carcinoma Adenoid cystic carcinoma Adenocarcinoma	50% of malignant salivary gland tumours Rare condition in children

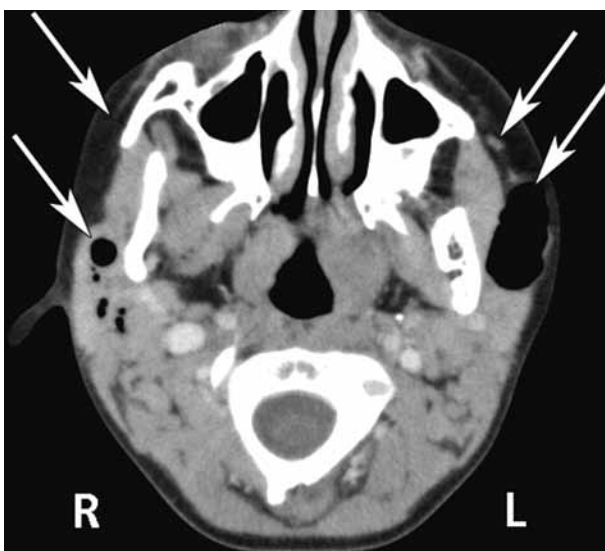


Figure 1

Air is clearly visible in both parotid glands, with dilated parotid ducts (arrows).

stressful situations. This maladaptive behaviour was explained to the parents and child in a reassuring manner and elimination of the precipitating manoeuvre resulted in resolution of the swelling. There was no need for further psychological support or care.

Discussion

Pneumoparotid is a benign condition of the parotid gland that involves air being trapped within the glandular ducts and parenchyma. Intra-oral pressure is normally between 2 and 3 mmHg; while, an increase in intra-oral pressure may lead to insufflation of air into

Table 2
Aetiology of pneumoparotid

Isolated event ^{1,4,9}	Dental procedures using air-powered instruments Forced exhalation during pulmonary function testing After general anaesthesia in patients receiving positive pressure Inflating a bicycle tire inner tube without a pump Blowing up a balloon Rapid decompression while scuba diving Severe coughing	Asthma COPD Attacks of hay fever
Recurrent episodes ^{1,8}	Professional habit	Players of wind instruments Glassblowers
Self-induced mechanism		Valsalva manoeuvre Psychiatric or psychosomatic disorders

Stensen's duct and the parotid gland. Under normal physiological conditions, the anatomical shape and localization of the orifice of Stensen's duct should prevent air insufflation. The duct is located lateral to the masseter muscle and enters into the oral cavity through the buccinator muscle. Contraction of the buccal muscles occurs when the cheeks are flattened and results in the duct being compressed. Furthermore, the orifice of the duct has a smaller diameter than the duct itself and it is slip-shaped in redundant mucosal folds.^{4,7,8} Table 2 gives an overview of the aetiology of pneumoparotid. A large group of patients with pneumoparotid have a psychiatric or psychosomatic background. Self-induced pneumoparotid has been described, especially in adolescents, to achieve prolonged school absenteeism or gain attention.^{7,9}

A symptom of pneumoparotid is parotid enlargement, this is usually unilateral, but it may also be bilateral. Palpation of the gland may produce crepitus when subcutaneous emphysema exists. Frothy saliva may appear from Stensen's duct intra-orally. The swelling of the parotid gland is not painful,

unless the gland is superinfected. Air can escape from the pneumoparotid parenchyma through the parotid capsule and subcutaneous emphysema appears. Progression of this process may lead to involvement of the parapharyngeal and retropharyngeal spaces; pneumomediastinum and pneumothorax have been described with potential airway obstruction.⁴ When prolonged dilation of Stensen's duct exists, there is a risk of sialiectasis formation with further ductal incompetence and insufflation. This condition may lead to predisposal to bacterial superinfections and recurrent or chronic parotitis.^{4,8}

Diagnosis is supported by accurate history taking and clinical observation. However, imaging is most helpful to reveal pneumoparotid and to assess its extension. Plain radiographs may reveal air in the parotid gland or duct, or in the subcutaneous tissue. Ultrasound may show hyperechoic areas, representing air in the parotid gland. Sialography may show dilation of Stensen's duct or the secondary ducts. Radiolucencies within the ducts may be seen and represent pockets of trapped air. Disadvantages of sialography include its invasiveness, the risk

of false negative results, and its contraindication in the presence of acute inflammation. A computerized tomography (CT) scan is the imaging technique of choice. Multislice CT can reveal very small amounts of air and the extension of the pathological process to surrounding tissues can be seen. MRI with T2-weighted images can perfectly show dilation of Stensen's duct and a loss of signal inside the parotid parenchyma due to insufflation of air. This examination is expensive and may not be suitable for children because the inspection time is relatively long.^{4,9}

Treatment of pneumoparotid depends on the cause. In cases of self-induced pneumoparotid, psychological counselling may be beneficial. Wind instrument players and glass blowers can be taught techniques to reduce the chance of air insufflation.⁹ Antibiotic treatment can be given in accidental cases to prevent secondary infection.¹ Supportive treatment can consist of massage of the parotid gland, use of nonsteroidal anti-inflammatory drugs, sialogogues, and adequate hydration. Surgical intervention may be appropriate when recurrent episodes of infection occur that do not respond to conservative measures. Transposition of Stensen's duct to the tonsillar fossa has been described; this technique is similar to that used in cases of uncontrolled drooling.⁴ Parotidectomy has been performed in exceptional cases.⁸

Conclusion

Pneumoparotid can lead to enlargement of the parotid region. The best way to visualize this condition is with CT imaging.

Management consists of reassurance and avoidance of activities which increase intraoral pressure. Pneumoparotid should be considered in the differential diagnosis of parotid gland enlargement in children and adolescents. Psychological factors that may contribute to this disease should be considered.

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References

1. Kirsch CM, Shinn J, Porzio R, Trefelner E, Kagawa FT, Wehner JH, Jensen WA. Pneumoparotid due to spirometry. *Chest*. 1999;116(5):1475-1478.
2. Maehara M, Ikeda K, Ohmura N, Sugimoto T, Harima K, Ino C, Sawada S. Multislice computer tomography of pneumoparotid: a case report. *Radiat Med*. 2005;23(2):147-150.
3. Balasubramanian S, Srinivas S, Aparna KR. Pneumoparotitis with subcutaneous emphysema. *Indian Pediatr*. 2008;45(1):58-60.
4. Luaces R, Ferreras J, Patiño B, Garcia-Rozado A, Vázquez I, López-Cedrún JL. Pneumoparotid: a case report and review of the literature. *J Oral Maxillofac Surg*. 2008;66(2):362-365.
5. Nahlieli O. Salivary gland inflammatory disorders in children. In: Kaban LB, Troulis MJ, Eds. *Pediatric Oral and Maxillofacial Surgery*. Saunders, Philadelphia, 2004:187-201.
6. Orvidas LJ, Kasperbauer JL, Lewis JE, Olsen KD, Lesnick TG. Pediatric parotid masses. *Arch Otolaryngol Head Neck Surg*. 2000;126(2):177-184.
7. Mandell DL. Disorders of the salivary glands in children. In: Myers EN, Ferris RL, Eds. *Salivary Gland Disorders*. Springer, Heidelberg, 2007:222-230.
8. Turkyilmaz Z, Karabulut R, Bayazit YA, Sonmez K, Koybasioglu A, Yilmaz M, Kemaloglu YK, Basaklar AC. Congenital neck masses in children and their embryologic and clinical features. *B-ENT*. 2008;4(1):7-18.
9. Rahbar R, Grimmer JF, Vargas SO, Robson CD, Mack JW, Perez-Atayde AR, Marcus KJ, Grier HE, Healy GB, McGill TJ. Mucoepidermoid carcinoma of the parotid gland in children: A 10-year experience. *Arch Otolaryngol Head Neck Surg*. 2006;132(4):375-380.

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