

Facial swelling caused by pneumoparotid: a case report and literature review

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Keywords

Pneumoparotitis, pneumoparotid, pneumoparotis, facial swelling, self-inflicted injury.

Abstract

Facial swelling in children is a common symptom in paediatric patients. Despite its frequent occurrence, searching for the right diagnosis can often be a challenge for the clinician. With this case report and review of the literature we intend to highlight a very specific cause of facial swelling: pneumoparotid. Pneumoparotid is described as the presence of air within the parotid system and can be complicated with subcutaneous emphysema and pneumomediastinum. We aim to make clinicians familiar with this syndrome, so they can recognize it in future patients and take the proper steps to diagnose and treat the patient.

Introduction

Facial swelling in children is a common symptom in paediatrics, and often a reason for parents to visit the emergency department. When a child presents with facial swelling, many physicians will quickly rule out an allergic reaction. Physicians will also take acute swelling caused by inflammation into consideration. History, clinical examination, medical imaging and the biochemical work-up will help the clinician differentiate between different causes of inflammation, such as lymphadenopathy, sinusitis, odontogenic infections and orbital / pre-septal cellulitis. It is also important to rule out renal causes of oedema. When these frequent aetiologies have been ruled out a broader differential diagnosis is needed (1). Knowledge of the facial anatomy, the different aetiologies and their clinical presentation is essential to differentiate between the several possible causes of facial swelling. The different causes of facial swelling can be divided into several groups. Firstly, there are possible allergic causes of acute facial swelling caused by food allergies, contact allergies or hereditary angio-oedema. Second there is acute facial swelling with infection or inflammation, caused by underlying lymphadenitis (viral or bacterial causes), sinusitis, odontogenic infections or orbital/pre-septal cellulitis. Thirdly, there is facial swelling in combination with generalised oedema caused by underlying renal or cardiac pathology. Fourth, there is the possibility of vascular malformations such as lymphangiomas and haemangiomas. If the swelling is not caused by any of the above, a clinician will consider tumoral lesions such as rhabdomyosarcoma, Langerhans cell histiocytosis, Ewing sarcoma, osteosarcoma and neuroblastoma.

Another important cause of facial swelling is pathology of the parotid gland. The parotid gland in itself can be the cause of unilateral or bilateral facial swelling. When one looks at parotid pathology in particular, the differential diagnosis of parotid swelling is very broad. These include infections, neoplasms, auto-immune pathology, iatrogenic causes and parotid duct pathology. With this case report and review of the literature we aim to highlight another very specific cause of parotid pathology: pneumoparotid.

Case report

A twelve year old boy presented to the emergency department with an acute exacerbation of facial pain and swelling. Initial swelling of the face started three days ago. There were no other significant elements in the patient's history, in particular no fever, no itch and no previous trauma. There were no respiratory or gastro-intestinal complaints. Diuresis was normal. He was fully vaccinated. One week prior to this presentation, he had already been admitted to the hospital for intravenous analgesia and antibiotics for a suspected bilateral parotitis following parotid duct infiltration.

Clinical examination revealed swelling of the face and neck and bilateral swelling of the eyelids. The overlying skin was intact and no lymphadenopathy was palpable. The patient did not have a rash or urticaria. Cardiorespiratory assessment was normal. There was no oedema of the extremities. Upon presentation, he was hemodynamically stable with no fever and no signs of anaphylaxis.

The initial differential diagnosis consisted of an allergic reaction with angio-oedema, generalised oedema caused by a nephrotic syndrome or cardiac pathology, parotid pathology and an EBV infection.

Laboratory examination showed a leucocyte count of $17.56 \times 10^9/L$ (ref range $4.50 - 13.00 \times 10^9/L$), neutrophil count $13.7 \times 10^9/L$ (ref range $1.8 - 8.0 \times 10^9/L$), CRP 0.8 mg/L (ref range < 5.0 mg/L) and amylase 120 U/L (ref range $28 - 100$ U/L). Urinalysis excluded a renal cause of the symptoms in the absence of proteinuria. The patient was admitted to the ward with a working diagnosis of recurrent parotitis and was treated with intravenous fluids, antibiotics and corticosteroids.

During his hospital stay, a more detailed study of his medical record revealed that he had been hospitalised nine times in different hospitals in the past year. He presented each time with bilateral swelling of the parotid glands and once with diffuse facial oedema. Each time he was treated with corticosteroids, antibiotics or analgesics. The working diagnosis at that point was a recurrent parotitis of childhood. Because of the extremely recurrent nature of the disease in this case, two-monthly infiltrations with corticosteroids in the parotid duct were performed, but this was insufficient to stop the episodes.

There were some elements in this case that challenged this working diagnosis. Firstly, this boy presented with a painful swelling of the face with bilateral oedema of the eyelids, a presentation that is not typical for a recurrent parotitis of childhood. Secondly, he did not respond well to the therapy that had been initiated. We therefore decided to consider a wider differential diagnosis of bilateral parotid swelling (see table 1) (2, 3).

Recurrent infections are not a likely diagnosis in our case as his white cell counts and acute phase reactants were within normal limits. Auto-immune pathology was excluded by consultation with the paediatric rheumatologist and biochemical testing. No ductal lithiasis or other types of obstruction were observed on the sialogram. We could exclude malignancies with radiological examinations. An iatrogenic cause was unlikely as he had not undergone any recent procedures. The only remaining diagnosis was pneumoparotid. A clinical re-evaluation revealed trismus, crepitus and pain on palpation of the swelling. A head CT showed discrete bilateral pneumoparotid and extensive subcutaneous emphysema in the head-neck area, extending into the parapharyngeal space, but also subcutaneously into the upper mediastinal space. The diagnosis of a pneumoparotid with subcutaneous emphysema and pneumomediastinum was made. When re-examining his past investigations we came upon one CT report from the previous year which already mentioned the term 'pneumoparotid', but apparently this diagnosis was not pursued. His other past investigations (sialography, two MRI's and ultrasounds) were all negative.

Extensive questioning could not reveal the underlying cause, but the recent loss of his father in combination with the loss of school days prompted us to consider an underlying psychological problem. Review of the literature showed that self-induced pneumoparotid has been described in adolescents with psychosocial issues. Extensive questioning could not reveal the mechanism of how the self-induction came about, but self-induced pneumoparotid can be subconscious. The boy was referred to a psychologist for further evaluation. Six months after the diagnosis, he still had symptoms twice a month. He was scheduled for an observation during a one week hospitalisation, followed by possible botox injections or ductal clipping to stop the flow of air through the duct. Unfortunately, the patient was lost to follow-up. Re-evaluation of his medical record revealed that he did not present to the emergency room with pneumoparotid again.

Literature search

We performed a systematic search of the literature for paediatric case

Table 1: Differential diagnosis of facial swelling and parotid pathology.

Facial swelling	Parotid pathology
Allergies <ul style="list-style-type: none"> - Food allergies - Contact allergies - Hereditary angio-oedema 	Neoplasms <ul style="list-style-type: none"> - Malignant - Benign
Inflammatory / infectious causes <ul style="list-style-type: none"> - Lymphadenitis (viral (EBV), bacterial) - Sinusitis - Odontogenic infections - Orbital / pre-septal cellulitis 	Infections <ul style="list-style-type: none"> - Bacterial sialadenitis - Viral (HIV, mumps) - Tuberculosis
Generalised oedema <ul style="list-style-type: none"> - Cardiac pathology - Nephrotic syndrome 	Auto-immune pathology <ul style="list-style-type: none"> - Recurrent parotitis of childhood - Sjögren disease - Granulomatous disease
Vascular malformations <ul style="list-style-type: none"> - Haemangioma - Lymphangioma 	Iatrogenic <ul style="list-style-type: none"> - Anaesthesia mumps - Iodide mumps - Radioactive iodine sialadenitis
Tumours <ul style="list-style-type: none"> - Rhabdomyosarcoma - Langerhans cell histiocytosis - Ewing sarcoma - Osteosarcoma - Neuroblastoma 	Parotid duct pathology <ul style="list-style-type: none"> - Sialolithiasis - Strictures - Pneumoparotid
Parotid pathology (cf. next column)	

Differential diagnosis of facial swelling and parotid pathology

Figure 1: Literature search and selected paediatric case reports.

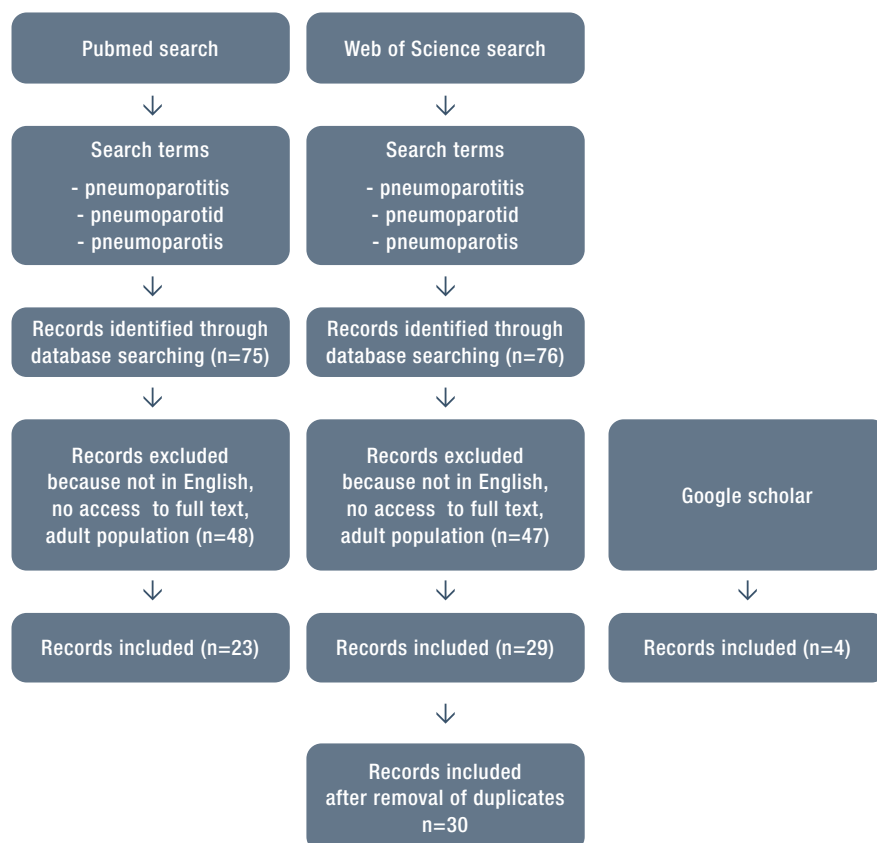


Table 2: Summary of the relevant clinical findings from the paediatric case reports.

Author	Date	Gender	Age	Complaints	Uni-bilateral	Recurrent	Investigations	Therapy	Cause
Ros et.al. (13)	1996	Boy	3 yrs	Pain, swelling	Unilateral	No	//	//	Blowing up balloons
Mutaf et.al.(14)	2006	Boy	3 yrs	Pain, swelling, redness	Unilateral	No	Blood test	NSAID	Anesthesia
Bowden et.al. (15)	2015	Boy	4 yrs	Pain, swelling, crepitus	Unilateral	No	X-ray, head CT	Reassurance	Blowing up balloons
Martin-Granizo et.al.(16)	1999	Girl	5 yrs	//	Uni- and bilateral	Yes	Blood test, X-ray, ultrasound, head CT	Antibiotics, hydration, warm compresses	//
Moschetta et.al. (8)	2021	Boy	5 yrs	Swelling, crepitus	Unilateral	No	Blood test, head CT	Antibiotics, anti-allergic therapy	Dental procedure
David et.al (17)	1988	Girl	6,5 yrs	Pain, swelling, warmth, redness	Unilateral	No	X-ray, MRI, head CT, cultures	Antibiotics, cannulating Stenson duct, positive reinforcement	Blowing the cheeks
Kyung et. al.(18)	2010	Girl	7 yrs	Pain, swelling	Bilateral	Yes	X-ray, head CT	Analgesia, antibiotics, stop trigger	Wind instrument player
McCormick et.al (10)	2012	Boy	7 yrs	Pain, swelling	Bilateral	Yes	Head CT	Antibiotics, NSAID, warm compresses, sialogogues	//
Martin-Granizo et.al. (16)	1999	Girl	8 yrs	Pain, swelling, crepitus	Unilateral	Yes	Sialography, head CT	//	//
Goguen et.al.(6)	1995	Girl	9 yrs	Pain, swelling, crepitus	Bilateral	Yes	Sialography, head CT	Antibiotics, massage, superficial parotidectomy	Valsalva manoeuvre
Gray et.al. (19)	2020	Girl	9 yrs	Swelling	Unilateral	No	Fine needle aspiration, head CT	//	Blowing the cheeks
Goguen et.al. (6)	1995	Boy	9 yrs	Swelling	Unilateral	Yes	Blood test, head CT	Antibiotics	Blowing against palm of hand
Krief et.al.(20)	1992	Boy	10 yrs	Pain, swelling, redness, crepitus	Bilateral		Blood test, fine needle aspiration, head CT, sialography	Antibiotics	Blowing the cheeks
Golz et. al.(21)	1999	Boy	10 yrs	Pain, swelling	Bilateral	Yes	Blood test, head CT	Psychological counseling	Blowing the cheeks
Luaces et. al.(22)	2006	Boy	11 yrs	Pain, swelling	Bilateral	Yes	X-ray, head CT	Antibiotics	Blowing the cheeks
Balasubramanian et.al. (7)	2007	Boy	11 yrs	Swelling, crepitus	Bilateral	Yes	Blood test, sialography, biopsy, head CT	Antibiotics	Valsalva manoeuvre
Lee et.al.(23)	2017	Boy	11 yrs	Pain, swelling, crepitus	Bilateral	No	Blood test, X-ray, ultrasound, nasolaryngoscopy, head CT	Antibiotics, analgesia, oxygen	
Markowitz-Spence et al. (24)	1987	Girl	12 yrs	Swelling	Bilateral	Yes	Blood test, sialography, head CT	Antibiotics, psychological evaluation	Valsalva manoeuvre
Grainger et al.(25)	2005	Girl	12 yrs	Pain, swelling	Bilateral	Yes	Sialography, head CT	Analgesia, antibiotics	Cold weather
Nassimbeni et al.(26)	1995	Boy	12 yrs	Swelling, crepitus	Bilateral	Yes	Blood test, sialography, head CT	Psychiatrist, psychologist	Wind instrument player, blowing the cheeks
Prabhu et al.(27)	2008	Boy	12 yrs		Bilateral	Yes	Head CT	Antibiotics	//
Ambrosino et. al. (11)	2019	Boy	12 yrs	Pain, swelling	Bilateral	Yes	Blood test, head CT, lip biopsy, sialendoscopy	Antibiotics, steroids, analgesia, massage	//
Rackowska-Labuda et.al. (28)	2019	Boy	12 yrs	Pain, swelling	Unilateral	Yes	Blood test, ultrasound, head CT, sialography	Antibiotics, intraductal dexamethasone	Blowing the cheeks
Aljeaid et.al.(29)	2020	Boy	12 yrs	Swelling	Unilateral	Yes	Blood test, head CT	Counseling	Blowing up balloons
Goguen et.al.(6)	1995	Boy	13 yrs	Swelling	Bilateral	Yes	Sialography, X-ray	Antibiotics, steroids, psychiatric counseling	Blowing the cheeks
Han et.al.(30)	2004	Boy	13 yrs	Swelling, crepitus	Uni- and bilateral	Yes	Sialography, head CT	Antibiotics, steroids, superficial parotidectomy, parotid duct ligation	//
Lagunas et.al.(9)	2017	Boy	13 yrs	Swelling	Bilateral	Yes	Head CT	Antibiotics, NSAID	Blowing the cheeks
Barros et.al. (31)	2022	Boy	13 yrs	Pain, swelling, crepitus	Bilateral	No	Blood test, ultrasound, head CT, X-ray	Antibiotics	Blowing up balloons
Sittel et.al.(32)	1999	Girl	14 yrs	Swelling	Bilateral	Yes	MRI	Antibiotics	Blowing the cheeks
Ferlito et.al.(33)	1992	Boy	14 yrs	Pain, swelling	Bilateral	Yes	Blood test, ultrasound, sialography, X-ray, fine needle aspiration	Antibiotics, NSAID, corticosteroids	Blowing the cheeks
Al Ohali et.al.(12)	2020	Boy	14 yrs	Swelling, crepitus	Unilateral		Head CT	Counseling	Blowing the cheeks
Birzgalis et.al.(34)	1993	Boy	16 yrs	Pain, swelling, crepitus	Unilateral	Yes	Blood test, head CT, fine needle aspiration	Antibiotics, parotidectomy	Valsalva manoeuvre
Alnaes et.al. (35)	2017	Girl	//	Pain, swelling, redness	Unilateral	Yes	Blood test, ultrasound, head CT, fine needle aspiration	Antibiotics	Blowing on a paper trumpet / sucking water bottles

reports of pneumoparotid. PubMed and Web of Science were searched on 09/12/2022 using the search terms 'pneumoparotid', 'pneumoparotitis' and 'pneumoparotis'. We retrieved all case reports in English, with full access texts and summarized the clinical findings in detail. We excluded 95 manuscripts based on age of the patients, language or full text access. After removal of duplicates, we reviewed 30 manuscripts from 1987 until present. Four case reports were selected after a search on Google Scholar (figure1). An overview of the relevant clinical findings from the paediatric case reports is shown in table 2.

Discussion and comparison with the international literature

Definition

Pneumoparotid, first described in 1865 by Hyrtl, is defined as 'the presence of air within the parotid system' (gland and Stenson duct) (4, 5). The earliest reports from pneumoparotid cases date from 1915 and involved soldiers from the French Foreign Legion in North Africa, who blew into a bottle to mimic mumps and as such avoid duty (2, 6). Pneumoparotid with associated infection is described as pneumoparotitis, and is a common complication (2, 4).

Mechanism

There are natural protective mechanisms that prevent air insufflation, but because of a supra-normal or sudden increase of intraoral pressure or defects in the preventive mechanisms of reflux, a retrograde insufflation of air through the Stenson duct can cause pneumoparotid (2, 5, 7, 8). Hypotonia of the buccinator muscle, hypertrophy of the masseter muscle and obstruction of the Stenson's duct by mucus are all risk factors (2, 4).

Cases of self-induced pneumoparotid are described in the literature (9).

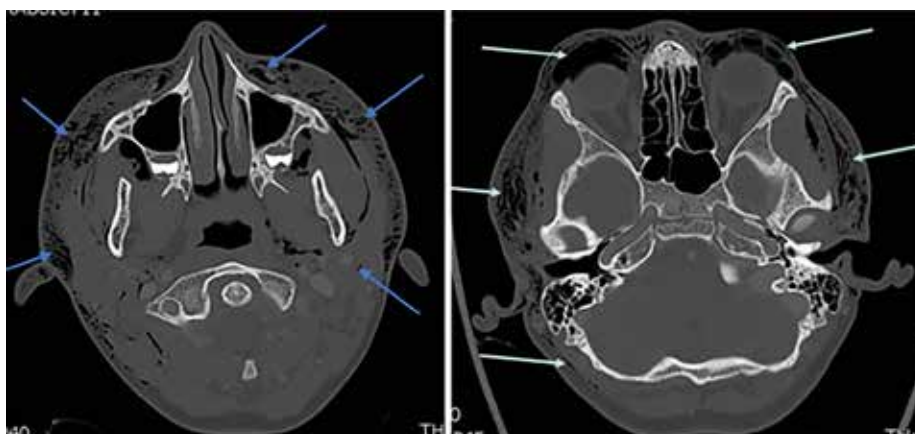
Pneumoparotid is often misdiagnosed as parotitis, because of the presence of oedema and local pain in the parotid gland (4).

In a few cases there is a presence of air in the parapharyngeal and retropharyngeal space, an uncommon complication. This happens when glandular acini break down because of high intra-oral pressure. This causes an air leak to the surrounding cervicofacial subcutaneous tissues and facilitates communication with the para- and retropharyngeal space. In these cases it is possible that the air extends to the mediastinum and causes a pneumothorax or a pneumomediastinum (2, 9).

Patient population

One can divide the patient population into four subgroups: occupational cases (wind instrumentalists, glass blowers, underwater divers), iatrogenic cases (anaesthesia, dental procedures, CPAP, spirometry), cases associated with inhalation (nitrous oxide, deodorants) and self-induced cases (Valsalva manoeuvres, aggressive nose blowing, chewing gum, chronic cough suppression) (2, 4, 6, 9). In our case we strongly suspect self-induced pneumoparotid, an entity that has been described in adolescents with psychosocial issues. It can be a subconscious or a deliberate attempt to gain secondary benefits (6). In the literature, cases of adolescents who have conflicts with

Figure 2: Head CT. Arrows = air.



parents, problems with adolescent adjustment, nervous tics, abnormal behaviour or previous psychiatric treatment are reported (7). Self-induced pneumoparotid has been associated with secondary benefits, such as school absenteeism, which should alert clinicians to look for a psychological cause (7, 9, 10).

Physical examination

Pneumoparotid is a rare condition and many patients with this symptom are misdiagnosed and therefore wrongly treated as a 'bilateral parotitis' or a 'recurrent parotitis of childhood'. These patients present with uni- or bilateral swelling at the site of the parotid gland (11). Some authors describe it as a painful swelling, others describe a pain-free swelling (9, 11). Sometimes there is erythema or subfebrillity and sometimes there is crepitus over the parotid area. In some patients air bubbles exude from the Stenson's duct during massage or palpation of the parotid gland (4, 6, 9, 10, 12). Pneumoparotid can predispose to sialectasis, bacterial parotitis, subcutaneous emphysema, pneumomediastinum and pneumothorax (4, 7).

Diagnosis

The gold standard for diagnosis is computed tomography (Fig 2.). The CT scan usually shows air in the parotid system and by extension air in the surrounding tissues, ductal dilation is also a common finding (2). Ultrasound is performed in many cases and is a useful tool to differentiate superficial swelling in the head-neck area. It shows multiple hyperechoic areas, corresponding to air in the glandular parenchyma (2). A sialography shows absence of stones in the parotid duct and can sometimes show a dilated Stenson duct or air within the ductal system (6, 9). Laboratory examination shows no evidence of systemic pathology. A slight elevation in the white blood cell count is possible, as well as an elevation in the amylase level. A biopsy of the gland is not necessary, but can show acute and chronic inflammatory changes in the parotid gland.

Treatment

The enlargement and swelling of the parotid resolves spontaneously over the course of a few days (2). In the literature there are suggestions to give prophylactic antibiotics to prevent pneumoparotitis, because of the increased risk of retrograde introduction of oral microbes in the gland (4). Other suggestions are the use of anti-inflammatory drugs, massaging the parotid, hydration, application of heat or sialagogues (2, 9, 10).

When one encounters a case of self-induced pneumoparotid it is important to educate the patient. Sometimes it is necessary to refer the patient for psychotherapy (2).

The last option for severe recurrent pneumoparotid is surgical treatment. Suggested treatment options are transposition of Stenson's duct to the tonsillar fossa, ligation of the Stenson's duct or superficial parotidectomy. Surgical treatment should only be considered in recurrent cases and when the quality of life is significantly impacted. (2, 9, 10).

Conclusion

Pneumoparotid is a phenomenon where there is air in the Stenson's duct and throughout the gland. In some cases there is subcutaneous emphysema and pneumomediastinum because of breakdown of the acini in the gland and presence of air in the parapharyngeal and retropharyngeal space. The most frequent cause in adults and children is self-induced pneumoparotid, most often because of frequently blowing up the cheeks. The gold standard for diagnosis is a cranial CT. There is no consensus in the literature regarding treatment. Numerous treatment options, such as antibiotics, corticosteroids, are already explored because of a delay in diagnosis. Self-induced pneumoparotid is best managed with psychological counselling to stop the behaviour. Sometimes the condition is complicated with an infection on the gland, namely pneumoparotitis. In that case, therapy with antibiotics is warranted. When there is a high recurrence rate, surgical treatment is an option.

Conflict of Interest Statement

The authors declare that there are no conflicts of interest with regards to the acquisition and reporting of the data of the study presented in this manuscript, all procedure were in line with the editorial policy of the Belgian Journal of Paediatrics.

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