■ CASE STUDIES

Parotid Pneumocele in Down's Syndrome

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Pneumoparotiditis is related to the cause of an infrequent increase in the size of the parotid gland due to the entrance of air through the Stenon duct following an increase in the pressure in the oral cavity. It is observed in musicians who play wind instruments, glass blowers, patients undergoing diagnostic or therapeutic techniques, in adolescents that provoke it intentionally, and in mentally handicapped people as found in the clinical case reported here. The symptoms are usually non-specific and the diagnosis is confirmed with an axial tomography. The interest in this clinical case lies in the peculiar form of presentation and its diagnostic confirmation through the use of CT imaging.

Key words: Parotid. Pneumoparotid. Pneumosialadenitis. Aerial parotiditis.

Neumocele parotídeo en el síndrome de Down

La neumoparotiditis supone una causa infrecuente de aumento de tamaño de la glándula parótida debido a la entrada de aire por el conducto de Stenon secundaria al incremento de presión en la cavidad oral. Se observa en músicos de instrumentos de viento, sopladores de vidrio, pacientes sometidos a técnicas diagnósticas y/o terapéuticas y, de forma intencionada, en adolescentes y disminuidos psíquicos, como en el caso clínico que presentamos. La clínica suele ser inespecífica y el diagnóstico se confirma mediante tomografía computarizada. El interés del caso clínico radica en la particular forma de presentación y la confirmación diagnóstica mediante la imagen de la tomografía.

Palabras clave: Parótida. Neumoparótida. Neumosialoadenitis. Parotitis aérea.

Pneumoparotiditis consists in the accumulation of air in the interior of the parotid gland due to an increase in intraoral pressure accompanied or not by an inflammatory process.[1]

It is characteristic of musicians playing wind instruments, glass blowers, etc. It can be observed in certain odontological[2] or spirometric[3] techniques, etc, in which the pressure inside the oral cavity is increased. In addition, self-induced cases have been observed in adolescents and the mentally handicapped.[4]

Pneumoceles of the salivary glands are not a common disorder, and frequently present as asymptomatic, except for the aesthetic effect caused by the tumour.

CASE STUDY

A 28-year-old woman with Down's syndrome, without other relevant history, attended an otorhinolaryngological clinic for an asymptomatic unilateral parotid tumour of rapid appearance together with superficial cutaneous lesions in the area that, according to relatives, were caused by the patient scratching herself. The only notable finding on examination was crackling on palpation of the gland. A simple fronto-occipital cranial radiography was performed (Figure 1) as well as computerized tomography of the neck with contrast (Figure 2), revealing a cavity filled with air in the interior of the parotid gland. After the performance of the imaging tests, evaluation of the patient was resumed, and the disappearance of the tumour was observed. Upon further questioning, the patient acknowledged that she

Figure 1. Simple radiography of the fronto-occipital cranium: hydroaerial accumulation projected over the left mandibular branch.

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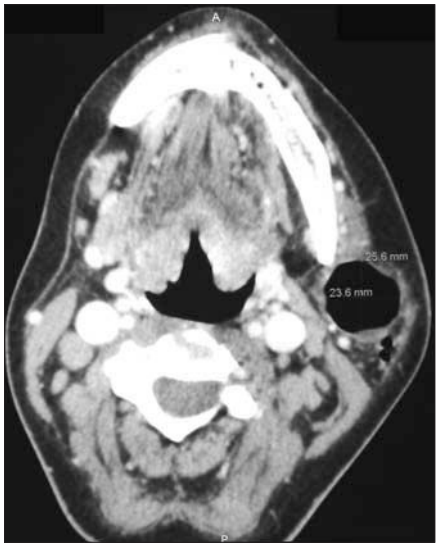

Figure 2. Computerized tomography of the neck with contrast: large amount of air identified in the parenchyma and the internal duct system of the left parotid gland. The largest accumulation measures 2.5 2.3 cm in cross-section.

provoked the insufflation of air within the gland by performing forced Valsalva techniques with the purpose of obtaining the attention and care of her parents and that later, through manual massage of the area, she reduces the swelling, which had returned to normal.

DISCUSSION

The parotid gland drains the interior of the oral cavity through the Stenon duct that, due to its anatomical characteristics (smaller diameter in the drainage ostium than in the rest of the duct, redundant mucous in the exit orifice, and the contraction of masseter and buccinator musculature collapsing it), prevents air, saliva, and/or bacteria reflux from the mouth towards the gland.[5]

The accumulation of air in the interior of the parotid gland has various names in the literature: pneumoparotitis, pneumoparotiditis, pneumoparotis, pneumocele of the parotid gland, aerial parotitis, pneumosialadenitis, etc.[6] The pathogenic mechanism is secondary to an increase in intraoral pressure and then, the entrance of air into the gland's interior. Intraoral pressure is 23 mm Hg in normal conditions and can reach 140-150 mm Hg in certain circumstances.[7]

The risk factors favouring this condition can be divided into occupational, as observed in wind instrument musicians, glass blowers, odontological, and spirometric techniques, following anaesthesia, after intubation with positive pressure, and in the practice of scuba diving; and non-occupational, in facial aggression, adolescents, and the mentally handicapped for attention-grabbing, Valsava techniques, etc.[1] Anomalies and malformations also exist at the level of the gland that may favour the development of pneumocele, such as ductal conditions, hypertrophy of the masseter muscle, or hypotonia of the buccinator.[6] The pneumocele may be unilateral or bilateral; it may manifest clinically with local symptoms such as pain, soft consistency and an increase in local temperature to the touch, subcutaneous emphysema with crackling along the Stenon duct (observed in 50% of cases) and/or bubbles emitted from the drainage orifice during massage of the gland, erythema, and fever. On many occasions there is an underlying condition of chronic parotiditis and sialadenitis.[8]

The spread of air from the parotid may produce subcutaneous emphysema in the face, neck, and mediastinum, and also pneumothorax.[9] The inflammatory reaction and the increase in amylase are due to dissection produced by the air in the glandular tissue.

The diagnosis is based on clinical history and physical examination of the patient together with certain imaging tests that serve to confirm the diagnosis, such as simple radiography of the head and neck that, in the acute phase, reveals air along the Stenon duct and in the interior of the gland. In severe cases, subcutaneous emphysema is observed; sialography may be normal or present dilatation of the duct; tomography reveals air in the parenchyma, the ductal system and the inside of the parotid gland.[6]

In the differential diagnosis, consideration has to be given to infectious processes (viral [the most frequent], bacterial, fungal, and microbacterial); autoimmune processes (Sjögren's syndrome, Wegener's vasculitis); malnutrition; endocrine conditions (diabetes, Cushing's disease, hypothyroidism); sarcoidosis; hepatic insufficiency; drugs; allergies; pregnancy; ductal obstruction; and benign or malignant neoplasias.[1]

The clinical symptoms tend to evolve satisfactorily and spontaneously, resolving itself in a matter of minutes or hours.

Treatment is individualized in each case. In the occupational forms, insufflation techniques should be reduced, applying behavioural correction methods, and psychiatric help should be provided in self-induced cases. Severe cases are treated conservatively through hydration, massage, local heat, and sialogogues; treatment with anti-inflammatory drugs and prophylactic antibiotherapy used to control pain and potential subsequent infection. Surgical treatment is reserved for chronic and/or recurring forms; a parotidectomy or ligature of the duct may be performed if appropriate.[1]

In spite of its scant incidence, in the face of a unilateral or bilateral parotid tumour with no specific symptoms, parotid pneumocele should be included among the possible differential diagnoses and special attention paid to clinical anamnesis to detect possible risk factors.

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