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CASE REPORT Open Access

Spontaneous regression of pseudotumor inflammatory parotitis: case presentation and literature review

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Abstract

Background: Pseudotumor inflammatory sialadenitis (PIS) is a rare disease. The differential diagnosis relays mainly on histopathology. We believe this is the first case of spontaneous regression of pseudotumor parotitis.

Case presentation: A 58-year-old male patient with a history of diabetes and hypertension presented a huge mass of the parotid gland suggesting an advanced stage malignancy. CT scan disclosed a large parotid tumor that invaded the parapharyngeal space and invaded the internal carotid artery in the parapharyngeal space. Complete surgical removal was therefore impossible. A biopsy was planted for further management according to the tumor board's recommendations. Within 5 weeks from the initial consultation, the tumor has completely and spontaneously resolved. The patient presented late to his biopsy appointment due to COVID free circulation restriction as he lived in another city. Furthermore, the workup ruled out autoimmune disease, viral-induced parotitis, inflammatory myofibroblastic tumor, and immunoglobulin G4— related inflammatory pseudotumor. Parotid gland biopsy revealed nonspecific fibrosis tissue. The patient remains free of disease without further treatment for a year follow-up.

Conclusions: Inflammatory pseudotumor parotitis is a manifestation of many conditions, few have been identified such as inflammatory myofibroblastic pseudotumor and immunoglobulin G4-related inflammatory pseudotumor but many remain unknown. Meticulous clinical and imaging, biology, immuno-pathology work-up is crucial for differential diagnosis.

Keywords: Inflammatory pseudotumor, Parotitis, Pseudotumor parotitis

Background

Pseudotumor inflammatory sialadenitis (PIS) is a rare condition that behaves clinically as salivary gland malignancy. Literature is scarce regarding this entity. The current trend is that it may be the manifestation of many conditions such as Inflammatory Myofibroblastic Tumor which was known before 1998 as inflammatory pseudotumor [1]. The latter can involve salivary glands, orbit, and abdominal organs and have a specific immunopathology profile. Another concept is "immunoglobulin G4-related inflammatory pseudotumor" [1, 2] which occurs in various anatomic sites including the liver, spleen, gastrointestinal tract, and lung and also may induce multifocal fibrosclerosis or chronic sclerosing sialadenitis. Although the etiology might not be found, the clinician has to rule out life-threatening conditions such as malignancy, autoimmune disease, and inflammatory conditions with specific serology or immunopathology profile. We believe this is the first case of spontaneous regression of pseudotumor inflammatory parotitis.

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Case presentation

A 58-year-old male patient with a history of diabetes and hypertension taking metformin and angiotensin receptor blockers, presented a rapidly growing mass on his right jaw that became stiff within two months associated with recent dysphagia and choking. There was no history of fever. The patient did not report any symptoms associated with impaired function of salivary or lacrimal glands. Physical examination found a huge stiff mass of the right parotid gland that involved also the submandibular region without invading the skin, no facial palsy or trismus were noticed. Also, the patient presented neither a scalp malignancy nor cervical lymphadenopathy. Nasofibroscopic examination of the upper aerodigestive tract has shown a significant bulging of the right lateral pharyngeal wall pushing the larynx to the left side with no compromised vocal folds movement. CT scan (Figs. 1 and 2) disclosed a large, smooth-surfaced, infiltrating mass of the right parotid gland, which invaded the parapharyngeal space and the internal carotid artery. This mass had a heterogeneous opaque shape after intravenous contrast injection. Complete surgical removal was therefore impossible. A biopsy was planted for further management according to the tumor board's recommendations. Within 5 weeks from the initial consultation, the tumor has completely and spontaneously resolved. The patient presented late to his biopsy appointment due to COVID free circulation restriction as he lived in another city. This remarking event in the course of this patient's pathology leads us to assume that it could be an atypical form of an auto-immune or inflammatory disease. The biology workup revealed a slightly elevated sedimentation rate with normal blood white cell count. No specific immunoglobulin profile on serum protein electrophoresis was found, autoimmune disease markers were negative, thoracic and abdominal CT scan was uneventful and viral serology was negative. A parotid gland biopsy was performed. Two specimens for pathological examination were collected, they measured approximately 15×6 mm and 20×6mm. They were fixed in 10% formaldehyde and embedded in paraffin for immunopathology study. The microscopic examination (Fig. 3) revealed fibrous tissue between salivary ducts without any atypical or inflammatory cells. In the salivary gland tissue, no mononuclear cells, or lymphocytes, and no evidence of malignancy was observed. The immunopathology study was negative for expressing anaplastic lymphoma kinase-1, CD-68, and muscle-specific actin. These markers are present in a variety of conditions such as anaplastic large cell lymphoma, malignant histiocytosis, histiocytic lymphoma, inflammatory myofibroblastic tumor, adenoid cystic carcinoma myoepithelial component, leiomyosarcoma, malignant fibrous histiocytoma, myoepithelioma, myofibroblastic sarcoma, myofibroblastoma, perivascular epithelioid cell tumors, pleomorphic adenoma, rhabdomyosarcoma,

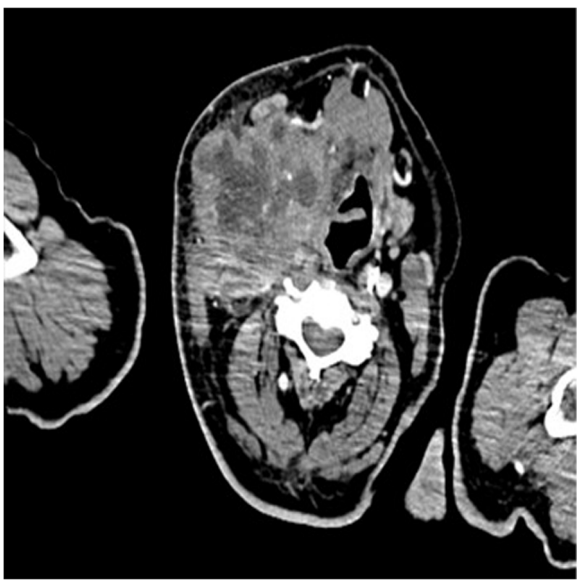

Fig. 1 CT scan axial view C+ showing a heterogeneous mass of the right parotid gland extended to the submandibular region and uncompressing the vascular axe with an important impact on the aerodigestive tract

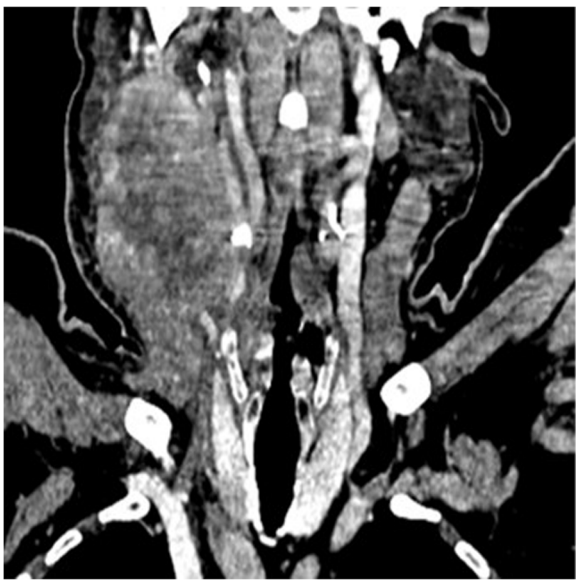

Fig. 2 CT scan coronal view C+ showing a heterogenous mass of the right parotid region uncompressing the vascular axe with no identified limit regarding the surrounding soft tissue

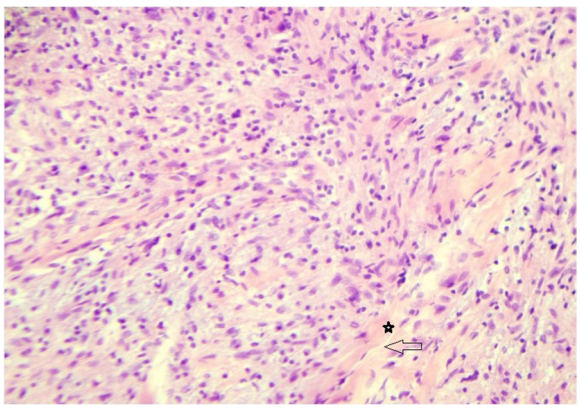

Fig. 3 Biopsy specimen of the parotid gland pathology features on hematoxylin–eosin stain showing normal glandular acini and fibrosis tissue between the salivary duct/400. The arrow points toward a salivary duct and the star mark fibrosis area

and solitary fibrous tumor. One year after spontaneous disease regression, the patient is clear of any symptoms, auto-immune disease markers and serology still negative at 3 months and six months of follow-up.

The patient's episodes of care are reported in Fig. 4.

Discussion

A PubMed literature search using MeSH "pseudotumor", "sialadenitis", "chronic", and "inflammatory" has identified only 24 articles, including mainly case reports and case series. The first case of inflammatory pseudotumor of the parotid gland was reported in 2004, authors described pathology features and insist on considering obstructive sialadenitis, nonspecific sialadenitis, and myoepithelioma as differential diagnoses [3]. PIS can be considered as a feature of a heterogeneous group of pathologies. This disease is rare and most commonly occurs in the submandibular gland [1–3]. The parotid gland is exceptionally affected. PIS can be an atypical manifestation of auto-immune diseases such as Gougerot-Sjogren syndrome [4] and rheumatoid arthritis [5]. Also, it may represent the first stage of chronic sclerosing sialadenitis which can be a manifestation of immunoglobulin G4-related inflammatory pseudotumors [4]. Symptoms usually lead clinicians to suspect a salivary gland neoplasm [6]. Possible differential diagnosis includes malignant tumors and other benign or inflammatory conditions of the salivary glands, such as inflammatory myofibroblastic tumors, granulomatous sialadenitis, radiation effects, and benign lymphoepithelial lesions and sialolithiasis, the latter is less common in the parotid gland. Chronic sclerosing sialadenitis is a benign inflammatory lesion of unknown etiology yet it has sometimes been associated with Ig G4-related inflammatory pseudotumors [4, 7]. Up until now, there have been no reports of malignancy of such a condition. In all cases reported in the literature, the diagnosis was set up after parotidectomy for well-circumscribed inflammatory lesion or tumor which can present central necrosis. No additional treatment was needed [8].

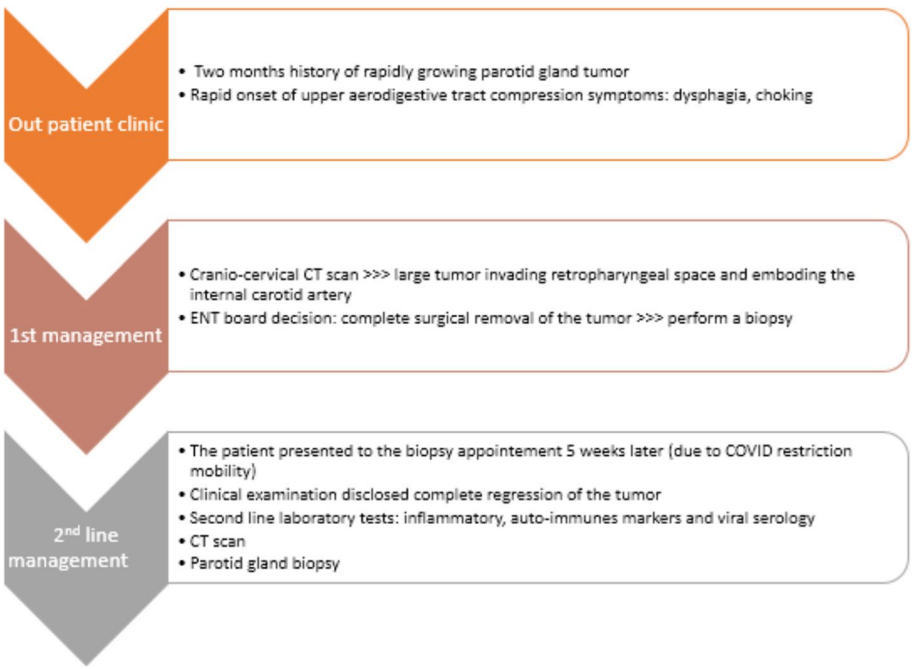

Fig. 4 The patient's episodes of care timeline

There is no agreement on the most suitable treatment for such a condition. Surgery is proposed bearing in mind differential diagnosis as histopathology study of the surgical specimen establishes the exact diagnosis. Complete removal of the tumor is performed whenever it is possible. Other therapeutic procedures reported in the literature [9, 10] are radiation, chemotherapy, azathioprine, indomethacin, and steroids. Steroids allow for reducing symptoms related to edema. However, their effect on the primary process is not proven. The particularity of our patient's case is the spontaneous tumor regression suddenly before the biopsy was performed, which did not help us establish an exact diagnosis. However, the existing fibrosis tissue in microscopic examination of PG sample and inflammatory markers in blood test were highly supporting the diagnosis of chronic sclerosing sialadenitis manifesting as a spontaneous regression of pseudotumor inflammatory parotitis. Auto-immune disease and other differential diagnoses were ruled out because of the history of the patient and the negative serology profile. The patient received no further treatment and had no evidence of recurrent disease for about 1 year of follow-up.

Conclusions

Pseudotumor inflammatory parotitis is a manifestation of many conditions. Few have been identified such as inflammatory myofibroblastic pseudotumor and immunoglobulin G4-related inflammatory pseudotumor but many remain unknown. Meticulous clinical and imaging, biology, and immunopathology work-up is crucial for differential diagnosis.

Patient's perspective

I initially had a swallowing and breathing difficulty with a rapidly growing mass in my cheek, two months later I consulted the doctor and performed a CT scan. Doctor announces to me that I have a tumor that cannot be removed completely and therefore I must have a biopsy for further consideration. I took some time to think about it. I took 3 weeks to a month then went back to my doctor for the biopsy. Surprisingly I was feeling better and noticed that I have no more symptoms. My doctor examined me and could not find the mass. So, after doctor counseling, she decided that I should have some blood tests and CT scans. Also, she insists on performing the biopsy to dismiss an unusual disease. Fortunately, all investigations went normal. I am always free of symptoms and beside my cardiac problems I am feeling very good and optimistic about the future. I have a medical follow-up every 6 months.

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Authors' contributions
NO was involved in diagnostic procedures and manuscript drafting, MA was involved in literature review and drafting of the manuscript, NH was involved in pathology study and reviewed the manuscript, and MNA reviewed the manuscript for insightful remarks. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets generated and/or analyzed during the current study are not publicly available due to patient's data confidentiality but are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
Not applicable.

Consent for publication
An informed consent for publication purposes was obtained from the patient. Written consent is available.

Competing interests
The authors declare that they have no competing interests.

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