

Surgical Management of Nonmalignant Parotid Masses in the Pediatric Population: The Montreal Children's Hospital's Experience

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Abstract

Nonmalignant parotid masses in children can have protean etiologies ranging from infective parotitis to a benign neoplastic, vascular, or congenital origin. We review the 10-year experience of a tertiary care pediatric centre with respect to the surgical management of nonmalignant parotid masses. In total, 15 patients with nonmalignant masses of the parotid gland region underwent surgery. Five children were diagnosed with lymphoepithelial cyst or first branchial cleft cyst. Three children were diagnosed with parotid abscess, one of whom had atypical mycobacteria. Other diagnoses included lymphangioma (three cases), chronic inflammation (two cases), and epidermoid cyst (one case). One patient who presented with a parotid cyst was diagnosed postoperatively with plexiform neurofibroma of the facial nerve. She was the only patient with postoperative facial nerve paresis, affecting the orbital branch. Presentation and postoperative complications of these surgically managed nonmalignant parotid masses are reviewed. The history and physical examination are of the utmost importance in predicting the diagnosis, although ultrasonography and computed tomography can be useful. Fine-needle aspiration cytology was not well tolerated by children and appears of little use as the accurate diagnosis was provided by the surgical pathology specimen.

Sommaire

L'étiologie des masses parotidiennes chez les enfants est très variée, depuis la parotidite infectieuse à la tumeur bénigne en passant par la lésion vasculaire ou congénitale. Nous avons donc revu notre expérience des 10 dernières années dans un centre de pédiatrie tertiaire pour ce qui est du management chirurgical des tumeurs bénignes de la parotide. Nous avons dénombré 15 patients qui ont subi une intervention chirurgicale à ce niveau. Cinq enfants avaient un diagnostic de kyste lymphoépithélial ou de kyste branchial; trois enfants avec des abcès dont un à mycobactérie atypique; et finalement trois lymphangiomes, deux inflammation chronique et un kyste épidermoïde. Un des enfants initialement considéré comme ayant un kyste parotidien a finalement été diagnostiqué comme porteur d'un neurofibrome plexiforme du nerf facial. C'est la seule enfant qui a développé une paralysie post-opératoire (branche orbitaire). Nous avons revu la présentation et les complications post-opératoires de ces masses traitées chirurgicalement. Ce sont l'histoire et l'examen physique qui sont les plus utiles pour prédire le diagnostic bien que l'échographie et la tomodensitométrie soient aussi utiles. La cytoponction n'a pas été bien tolérée par les enfants et n'apparaît pas d'une grande utilité puisque le diagnostic est donné de toute façon par le spécimen pathologique au moment de la chirurgie.

Key words: branchial cleft cysts, lymphangioma, nonmalignant, parotid masses, pediatric, plexiform neurofibroma

Parotid swelling in children is usually secondary to infective parotitis and rarely requires surgical intervention. Other etiologies of parotid masses include neoplastic, vascular, or congenital origins. The exact

nature of a mass in the parotid area can be difficult to ascertain, and the final diagnosis is often made by the pathologist.¹

Although the literature presents ample information on cancer of the parotid gland, nonmalignant parotid tumours are underdiscussed,² especially in the pediatric population. This article reviews the 10-year experience of a tertiary care pediatric centre with respect to surgical management of nonmalignant parotid masses.

Methods and Results

We reviewed our experience from 1986 to 1996 with patients presenting to the Montreal Children's Hospital with a parotid mass. We selected all patients who

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were surgically treated for a nonmalignant parotid mass. We present the demographic data, clinical presentations, pathologic findings, investigations, and treatment modalities.

In total, 15 patients with nonmalignant masses of the parotid gland underwent surgery. The age range was from 7 months to 18 years, with an average age at presentation of 7 years. There were 8 females and 7 males. The distribution of the final diagnosis is outlined in Figure 1.

Five children were diagnosed with lymphoepithelial cyst or first branchial cleft cyst. There were four girls and one boy. Clinical presentations consisted mainly of parotid swelling; one patient had bilateral preauricular sinuses, and another had a fistula to the external auditory canal. Three patients had a computed tomographic (CT) scan preoperatively; the others were diagnosed clinically. Surgery consisted of cyst excision, with superficial parotidectomy required in three cases.

Three children presented with an acute tender parotid swelling and were febrile. They were diagnosed with parotid abscess based on their clinical picture and ultrasound imaging. All patients underwent incision and drainage of their abscess and were put on broad-spectrum antimicrobial agents. Microbiology analysis of the draining fluid revealed *Staphylococcus aureus* in one patient and *Streptococcus hemolyticus* in another. The third patient is interesting in that unlike the two previous children, he had recurrence of parotid swelling after the incision and drainage. Bacteriologic studies demonstrated atypical mycobacteria. He was later found to have a positive purified protein derivative test.

All three patients were followed for 1 year without any complications. Two children had a history of chronic recurrent swelling of the parotid area. They underwent a superficial parotidectomy. Pathology

showed a picture of chronic inflammation. No complications were recorded at 1-year follow-up.

Three children were diagnosed with lymphangioma on the basis of either ultrasonography or CT. Two had a superficial and one had a total parotidectomy because of the deep extension of his lesion. Despite meticulous efforts to remove it completely, it was felt postoperatively that part of the mass was left behind.

One patient had an epidermoid cyst, diagnosed following the excision of a cystic lesion demonstrated in the right parotid on ultrasonography. Another patient who also presented with a parotid cyst confirmed on CT scan and magnetic resonance imaging was diagnosed postoperatively with plexiform neurofibroma of the facial nerve. She was left with a weak orbital nerve branch.

Discussion

Although 5% of all salivary gland neoplasms occur in children,³⁻⁵ these have been the focus of the medical literature, with much less emphasis placed on non-neoplastic processes affecting the parotid area (Table 1). Yet these can present both diagnostic and management challenges to the head and neck surgeon.

Our series addresses nonmalignant parotid masses requiring surgical intervention. None of the cases of parotid hemangiomas required surgical excision, and, surprisingly, we did not have any case of pleomorphic adenoma. In our data, the most common etiology of nonmalignant masses in the parotid area was of congenital origin, namely, first branchial arch anomalies followed by lymphangioma and dermoid cyst. First branchial cleft cysts or parotid lymphoepithelial cysts are rare, making up less than 10% of all branchial cleft anomalies.^{1,6,7} The latter stem from incomplete closure of the ectodermal portion of the first branchial cleft. It is the degree of closure that determines whether the anomaly will be a cyst, a sinus, or a fistula. These anomalies can have protean presentations but usually are associated with infection.⁸ Classic history includes multiple previous incision and drainage procedures for recurrent cystic masses.⁹ They are most frequently located close to the parotid gland, especially the superficial lobe that overlies the lesion. In the series of Triglia et al.,⁸ 35% of patients with first branchial cleft anomalies had parotid symptoms. Complete cure requires aggressive surgical excision and removal of the entire lesion. One of five patients in our series had a recurrence after simple excision requiring superficial parotidectomy to achieve a permanent cure. About 50% of the patients in the series of Triglia et al. had previous unsuccessful treatment. Work has divided these lesions into two types: type I is purely of ectodermal origin, whereas type II has both ectodermal and mesodermal origins.⁸

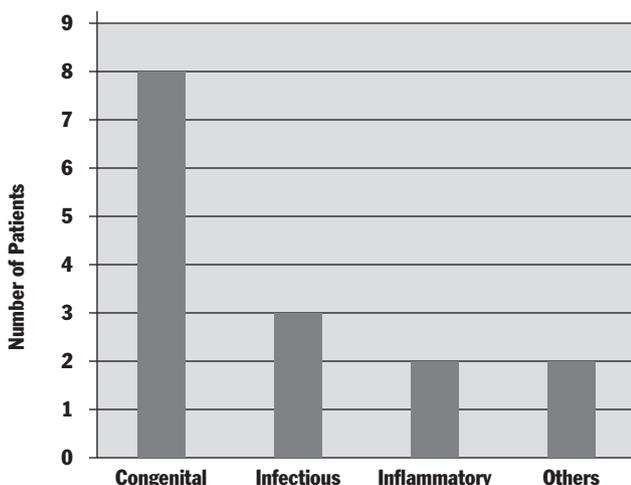


Figure 1 Distribution of etiologies of nonmalignant parotid gland masses in this series.

Table 1 Etiologies of Non-neoplastic Parotid Area Masses in 15 Children.

Infectious
Acute
Viral
Bacterial
Atypical organisms
Chronic
Congenital
First branchial arch anomalies
Hemangioma
Lymphangioma
Dermoid cyst
Neoplastic
Pleomorphic adenoma
Intraparotid facial nerve neurofibroma
Intraparotid facial nerve schwannoma
Others
Lipomatosis

Parotid lymphangioma is very rare. It is usually detected in infancy or early childhood. Eighty to 90% of patients are diagnosed by the second year of life. The children in our series were unusual in that they were diagnosed at the ages of 7, 9, and 12 years. Lymphangiomas grow slowly and present as painless, fluctuant soft masses.¹⁰ The diagnosis can be made by ultrasonography or CT.¹¹

One patient had a dermoid cyst diagnosed on pathology. These usually present as painless masses that are superficial or deep within the substance of the parotid gland and can be closely associated with the glandular parenchyma.

The next most common diagnosis in our series is parotitis. The latter can be classified into three categories: infectious, juvenile recurrent parotitis, and chronic. Infectious parotitis can be viral in origin. The most common pathogens are paramyxovirus, Epstein-Barr virus, coxsackievirus, influenza A, and parainfluenza viruses. Bacterial infection can be associated with acute and recurrent suppurative parotitis or intraparotid abscess. Juvenile recurrent parotitis is associated with reversible radiographic finding of sialectasis. Chronic parotitis usually presents with persistent or intermittent pain and swelling in the absence of infectious etiology or sepsis. Chronic inflammation is demonstrated on pathology.

Three of our patients had a parotid abscess. Abscess formation is thought to arise from primary parenchymal infection, ductal ectasis, or suppurative infection of subcapsular lymph nodes. Normal Stensen's duct secretion does not rule out the presence of an abscess. The most common bacterial pathogens are *S. aureus* and *Streptococcus* species. Occasionally, gram-negative bacteria and anaerobic species have been incriminated. Antimicrobial agents used should be resistant to β -lactamase because the latter is produced by 75% of pathogens. One of the most infrequent

causes of parotitis, present in one of our patients, is atypical *Mycobacterium tuberculosis*. When the diagnosis is made preoperatively, some cases respond to antimicrobial agents such as clarithromycin, isoniazid, rifampin, pyrazinamide, and ethambutol. Multiple agents are more effective than single-agent therapy. Unfortunately, the diagnosis is often made postoperatively—hence the importance of clinical suspicion. Left untreated, this condition can lead to central caseation and the development of fistulae and fibrosis.

Neoplasm of the facial nerve presenting as a parotid mass is extremely uncommon. The neural etiology is diagnosed intraoperatively in the absence of facial signs. The majority of these neurogenic neoplasms are schwannomas. We report the first case of intraparotid facial nerve neurofibroma in a pediatric patient. This was the only patient in this series who had postoperative facial nerve involvement affecting the orbital branch. This did not surprise us considering the nature of the tumour. The patient still had some motion left probably because of cross-innervation.

We did not find fine-needle aspiration useful in our pediatric population. In general, the procedure is poorly tolerated by children, and accurate diagnosis depends on a reliable, experienced cytopathologist.

Conclusion

The parotid glands can be subject to many nonmalignant pathologic processes that require surgical treatment. It is our impression that nonmalignant lesions are under-reported in the literature. These include, among others, branchial cleft cysts, lymphangiomas, parotitis, dermoid cysts, and facial nerve tumours. The history and physical examination are of the utmost importance. Ultrasonography and CT can be useful tools.

Although fine-needle aspiration cytology can occasionally help with the diagnosis, particularly in distinguishing tumours from inflammatory disease, surgical excision is often warranted. The final diagnosis is usually made on the surgical pathology specimen.

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