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CASE REPORT Open Access

Giant pediatric de novo parapharyngeal space pleomorphic adenoma: excision by semi trans-parotid cervical approach

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Abstract

Background Parapharyngeal Space tumors are rare tumors of the head and neck. They most commonly arise from the deep lobe of the parotid gland. Most of these tumors are in seen in adults and de novo origin from the minor salivary glands is extremely rare. The surgical approach needs to tailor made depending on the extent of the disease.

Case presentation A female child presented with chief complaints of swelling below the left ear for 9 months. On examination, the swelling was firm and non-tender, measuring 6 × 4 cm. A smooth bulge was seen in the left oropharynx, pushing the uvula to the opposite side. Fine Needle Aspiration Cytology (FNAC) showed features of Pleomorphic Adenoma. Contrast-Enhanced Magnetic Resonance Imaging (MRI) revealed a 4 × 6.1 × 6.7 cm well-defined solid lesion in the left parapharyngeal space. It was isointense on T1 and hyperintense on T2. Surgical excision was done via semi trans-parotid cervical approach. The tumor was solid and seen separately from the parotid gland, suggestive of a de novo tumor diagnosed as pleomorphic adenoma on histopathology. The patient made a good recovery and is disease free at 1 year of follow-up.

Conclusion Semi-transparotid combined with the cervical approach is a feasible option for large parapharyngeal space tumors without the need for mandibulotomy.

Keywords Parapharyngeal space tumors, Pediatric tumors

Background

Parapharyngeal Space (PPS) lesions account for only 0.5 to 1% of all head and neck tumors. The majority of these (80%) are benign. The most common PPS tumor is a pleomorphic adenoma, which originates from the parotid gland's deep lobe. Very rarely, these tumors can also arise from the aberrant minor salivary glands of the PPS [1].

The pyramid-shaped PPS extends from the skull base to the hyoid. The fascia from the styloid process to tensor veli palatini divides it into pre-styloid (anterior) and post-styloid (posterior) compartments. The pre-styloid compartment consists of the deep lobe of the parotid gland, lymph nodes and fat, whereas the post-styloid compartment has crucial neurovascular structures, including the internal jugular vein, internal carotid artery, cervical sympathetic chain, and cranial nerves IX, X, XI, and XII [2].

The pre-styloid compartment most commonly harbors salivary gland lesions, lipomas or lymphomas. The post-styloid compartment pathologies include paragangliomas, schwannomas, and nerve sheath tumors. PPS tumors commonly present with intraoral or neck swelling. FNAC and imaging (CT and MRI) remain the gold standard for diagnosis. Surgery is the mainstay of treatment with various approaches, the most common being transoral, transcervical, and transparotid [3].

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De-novo PPS pleomorphic adenoma is a rare entity, and very few case reports exist in today's literature. We here report a case of a giant pediatric de-novo pleomorphic adenoma that was managed surgically via semi trans-parotid cervical approach without the need for mandibulotomy. We also review existing literature on common presentations, investigations, and various surgical approaches, with their merits and demerits.

Case presentation

A female child from the Uttarakhand state of India presented with a swelling below the left ear for the past 9 months. It was initially the size of a pea and gradually progressed to its current size. However, the swelling was not associated with pain, dysphagia, or respiratory distress. The family history and the past history were unremarkable.

On local examination, a smooth, firm, round swelling measuring 6 × 4 cm was present below the left ear. It was extending from the tragus superiorly to 2 cm below the angle of the mandible inferiorly. The posterior extent was till the anterior border of the left sternocleidomastoid. It was non-tender and not fluctuant. A smooth bulge was visualized on the left side of the oropharynx, pushing the uvula to the right. However, the mouth opening was adequate (admitting three fingers). General physical and systemic examination was normal (Figs. 1A and 2A).

After the suspicion of a PPS tumor based on history and examination, the patient underwent CEMRI and FNAC to characterize the nature and extent of the lesion. Contrast-Enhanced MRI showed a well-defined

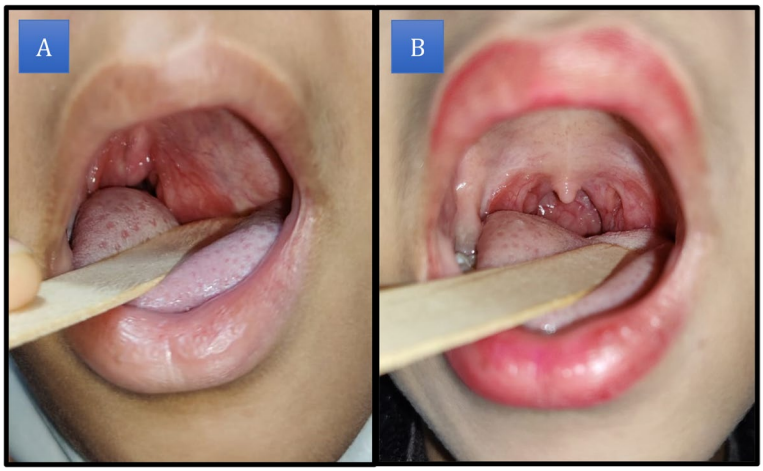

Fig. 1 Oral examination showing buldge in the left oropharynx preoperatively **A**, and normal postoperative picture **B**

Fig. 2 Neck examination showing swelling below the left ear preoperatively **A**, and scar three weeks postoperatively **B**

solid lesion measuring 6.7 × 6.1 × 4 cm in the left PPS, which was hyperintense on T2 and isointense on T1. It was extending into the masticator space and displacing the submandibular gland inferiorly. It was pushing the tonsil medially, narrowing the oropharyngeal airway and displacing the parotid gland laterally.

The tumor was seen separately from the deep lobe of the parotid gland by a fine translucent line of fibrofatty tissue (Fig. 3). FNAC from the swelling showed clusters of oval to spindle-shaped epithelial cells with scattered myoepithelial cells, suggestive of pleomorphic adenoma.

The MRI findings were consistent with the pre-styloid PPS tumor since it was pushing the internal carotid artery posteriorly. Benign neoplasm of the parotid gland is hyperintense on T2W, in contrast to malignant lesions, which are hypointense on T2W [4]. Since, this lesion was well defined and T2 hyperintense, a benign salivary gland PPS tumor was kept as a provisional diagnosis, which was further confirmed with FNAC.

With the left PPS Pleomorphic Adenoma diagnosis, excision was planned via a combined semi transparotid and transcervical approach under general anesthesia. The nature of the disease, its sequelae, management options, benefits of surgery and complications were discussed with the parents since the patient was a minor. After obtaining the written informed consent, the patient was taken up for surgery.

The tumor was approached via modified Blair's incision. Dissection proceeded in the subplatysmal plane and along Superficial Musculo Aponeurotic System. After separating the tumor from the sternocleidomastoid muscle, the marginal mandibular nerve was identified at the lower border of the mandible. This was followed in a retrograde direction until the facial nerve's main trunk (Fig. 4). The upper cervical nerve branch was also identified during the dissection and preserved. The superficial lobe of the parotid was dissected off the deep lobe only along the lower two divisions of the facial nerve to ensure the preservation of the parotid tissue. The lower part of the superficial lobe of the parotid gland was raised as a flap for medial exposure. The tumor was located deeper to the deep lobe of the parotid with a distinct plane in between. The tumor was abutting the digastric muscle and was deep to it. The digastric was first retracted inferiorly but the adequate exposure could not be achieved. The posterior belly was then divided in its mid portion to gain exposure to the parapharyngeal space. Blunt dissection was done around the tumor to free it from surrounding attachments while preserving the neurovascular structures (Fig. 5). The tumor was removed in toto. The cut

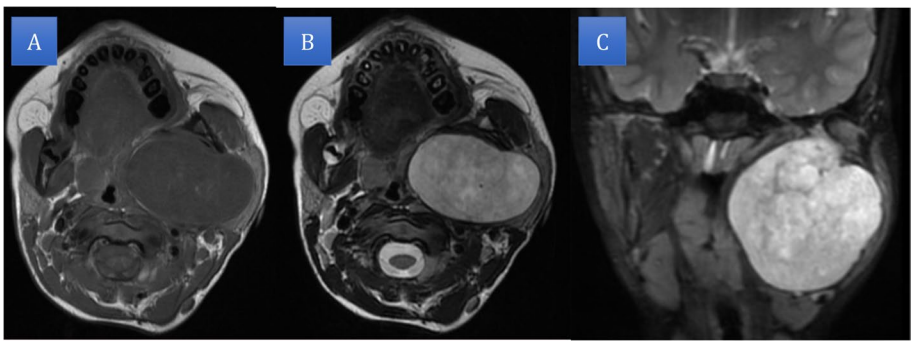

Fig. 3 Contrast-enhanced MRI showing the tumor to be well defined, T1 isointense **A** and T2 hyperintense **B**, **C**

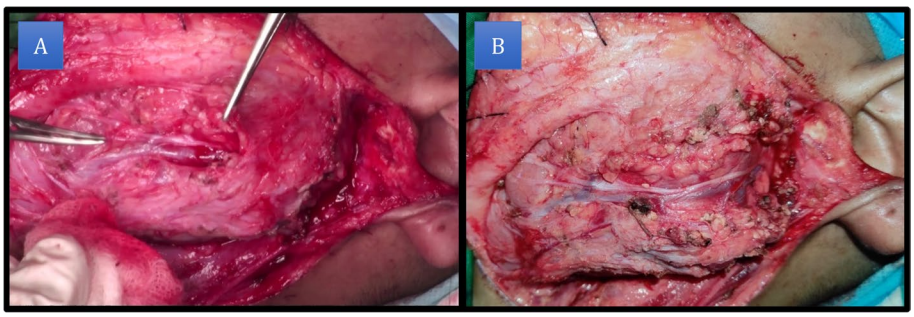

Fig. 4 Retrograde dissection of the facial nerve

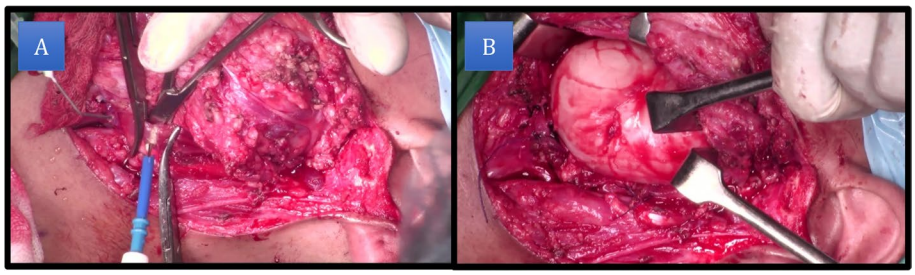

Fig. 5 **A** Posterior belly of digastric being divided, and **B** tumor exposed and seen separate from the deep lobe of the parotid gland, suggestive of a de-novo pleomorphic adenoma

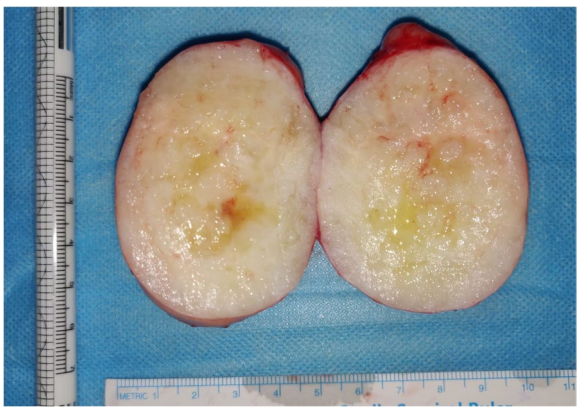

Fig. 6 Gross morphology of the excised tumor. A well-encapsulated globular tissue measuring 6 × 5.5 × 4.5 cm with homogenous grey white stroma on the cut section

ends of posterior belly of digastric were sutured. The raised portion of superficial lobe of parotid gland was reposited back and was sutured to the sternocleidomastoid muscle to bury the exposed facial nerve branches. The drain was placed in the postoperative cavity, which was removed on the 2nd postoperative day.

The postoperative period was uneventful. Sutures were removed on the 7th postoperative day, and the wound was healthy. The patient developed left marginal mandibular nerve paresis, which resolved completely 4 weeks postoperatively. The patient was discharged on the 8th postoperative day.

Postoperative histopathological examination showed a 6 × 5.4 × 4.4 cm well-encapsulated globular soft tissue, which was homogenous and grey in color (Fig. 6). Along with this, a reactive parotid lymph node was also identified. The patient was able to resume school 2 weeks after the surgery. On reviewing the patient 1 month after the surgery, palatal paresis and marginal mandibular nerve paresis resolved completely. The patient was followed up every 3 months thereafter. The patient completed 1 year of follow-up after the surgery with no residual or recurrence. The intra-oral buldge also subsided. The wound site was healthy, and there were no neurological deficits (Figs. 1B and 2B).

Discussion

Aetiology

In a systematic review by Riffat et al. (2014), 918 of 1118 PPS tumors were benign, 34% of which were pleomorphic adenoma. However, no case of de-novo minor salivary gland pleomorphic adenoma was reported [5].

Minor salivary tumors contribute to only 10% of all salivary gland neoplasms. However, 40% of these tumors are malignant. The palate was the most common site, followed by lips and buccal mucosa, with PPS being one of the least common sites [6].

A literature search was done on "PubMed" and "Google Scholar" using the keywords "parapharyngeal space, "minor salivary gland", and "pleomorphic adenoma". Case reports and series of de novo PPS pleomorphic adenoma were selected. A total of 17 articles with 19 cases of de novo PPS pleomorphic adenoma were found, out of which only 2 case reports were found in the pediatric population. The case report by Bhadani et al. of a 15-year-old female child does not mention the approach used for excision [7]. The other report by Brigger et al. of a 16-year-old male child is a small tumor measuring 4.5 × 3.5 × 3.0 cm excised transorally [8]. Our case is the 20th case reported to date. However, it is the third case reported in the pediatric population and the youngest age at which this disease has been reported (12 years). The tumor size in our case is the largest (measuring 6.0 × 5.5 × 4.5 cm) in this age group (Table 1).

Clinical presentation

PPS tumors can present with a broad spectrum of symptoms, from being asymptomatic to presenting with life-threatening cranial nerve palsies. The first and the most common presentation is neck swelling [22, 23]. Other presentations may include dysphagia, dysphonia, otitis media secondary to eustachian tube dysfunction and cranial nerve palsy. The most

Table 1 Literature review, demography, tumor size, and surgical approaches of all reported de novo PPS pleomorphic adenoma (in chronological order)

S. No	Author	Year	Age	Sex	Tumor size (cm)	Surgical approach
1	Bent et al. [9]	1992	35	Female	4.4 × 2.8	Transoral
2	Varghese et al. [10]	2003	40	Male	6.0 × 4.0 × 2.5	Mandibular swing
3	Brigger et al. [8]	2005	16	Male	4.5 × 3.5 × 3.0	Transoral
4	Hakeem et al. [1]	2009	20	Male	8.0 × 6.0	Transcervical
5	Hakeem et al.[1]	2009	53	Male	6.0 × 5.0	Transcervical
6	Rawat et al. [11]	2012	42	Female	8.0 × 7.0 × 5.0	Mandibular swing
7	Bhadani et al. [7]	2013	15	Female	7.5 × 5.5 × 4.5	NA
8	Jain et al. [12]	2013	35	Female	4.8 × 3.1 × 5.0	Transoral
9	Hwang et al. [13]	2013	34	Male	8.4 × 6.5 × 3.9	Transoral – transcervical
10	Akin et al. [14]	2014	50	Male	9.0 × 6.0 × 2.5	Transcervical
11	Akin et al. [14]	2014	56	Male	3.0 × 4.0	Transcervical
12	Phookan et al. [6]	2015	45	Female	8.0 × 6.0 × 5.0	Mandibular swing
13	Ray et al. [15]	2016	54	Female	6.0 × 6.0	Transcervical
14	Laturiya et al. [16]	2016	27	Female	6.5 × 5.5 × 4.2	Transcervical-transoral
15	Bist et al. [17]	2016	60	Male	10 × 8.2 × 5.8	Transcervical
16	Malhotra et al. [18]	2017	25	Male	13 × 15 × 8.0	Transcervical
17	Sagar et al. [19]	2018	25	Male	15 × 11 × 7.5	Transcervical-transparotid
18	Ahmed A et al. [20]	2019	44	Female	5.1 × 3.4 × 4.4	Transcervical
19	Galleti et al. [21]	2019	63	Male	8.0 × 6.0	Transcervical
20	**Present study**	**2022**	**12**	**Female**	**6.0 × 5.5 × 4.5**	**Transcervical-transparotid**

common presentation in a review of 537 cases by Riffat et al. was neck swelling (50%) and intraoral swelling (44%) [5].

Investigations

The ultrasound-guided cytology and imaging (CT and MRI) are invaluable diagnostic tools that complement each other to define the nature and extent of the disease. The direction of PPS fat displacement can help locate the tumor's origin. The internal carotid artery is displaced posteriorly by pre-styloid lesions and anteromedially by post-styloid lesions. Extra parotid lesions will demonstrate the presence of a fat plane between the deep lobe and the tumor [9].

Regular margins and maintained fat planes on CEMRI point to a benign pathology. CEMRI is considered more sensitive than computed tomography in determining the tumor's nature, extent, and relation to the neighboring structures [10].

Incisional biopsy is no longer indicated due to the anticipated risk of tumor seeding and subsequent recurrence. Hence, the gold standard for histopathological diagnosis is FNAC (Fine Needle Aspiration Cytology) [11]. FNAC is highly sensitive (0.80) and specific (0.97) for salivary gland neoplasms, but the reports can be inconclusive in about 6% of the cases [12–14].

Surgical approaches

An ideal surgical approach should provide maximum exposure, minimal morbidity, and the best possible cosmesis. Various approaches to the PPS have been described in the literature, each with its own merits and demerits.

Ashraf Hamza [15] and Malone et al. [16] have described the resection of the PPS tumors (90 to 100%) using the trans-cervical approach. A combination of trans-cervical with trans-parotid approach was described by Hughes et al. in 172 patients [17]. Trans-oral approach was described in the 1950s by Ehrlich [18]. This avoids the neck incision and its associated complications. It also provides direct access to the PPS, avoiding injury to the facial nerve (vide trans-parotid approaches). This comes at the cost of poor vascular control. McElroth et al. took vascular control with this approach in his series of 112 patients [1].

Malone et al. [16] (2001) found the transcervical approach to give adequate vascular control and tumor exposure in his series of 33 PPS tumors, with minimal need for tracheostomy, mandibulotomy, or facial nerve dissection [16]. Transcervical approach was found to be the first choice in PPS tumors by Chang et al. (2012), Presutti et al. (2012), and Basaran et al. (2014) [19–21].

Though mandibulotomy provides direct access to the PPS, it is associated with significantly morbid

complications such as non-union, mal-union, difficulty in mastication and facial asymmetry. Such complications can significantly affect an individual's quality of life and are highly undesirable, more so for benign pathology.

The transcervical approach can also be extended to a combined transcervical-transparotid approach. The largest de-novo PPS tumor excised via trans-cervical approach was by Malhotra et al. ($13 \times 15 \times 8$ cm, weighing 420 g) [24].

Olsen recommends a transcervical-transparotid approach for tumors of the minor salivary gland and the deep lobe of the parotid. He also advocated that the exposure can be increased by dividing the styloid process [2].

Hwang et al. [25] (2013) reported the excision of a large de-novo PPS pleomorphic adenoma ($8.4 \times 6.5 \times 3.9$ cm) by combining trans-oral and trans-cervical approaches. However, in this case, the patient developed a deep neck space abscess 1 week after the surgery, which had to be drained surgically. This perhaps also brings to light the risk of infection in combining trans-oral with the transcervical approach, which can be fatal with the exposed great vessels.

The tumor in our case measured $6 \times 5.5 \times 4.5$ cm in a 12-year-old child. To the best of our knowledge, such a large de-novo PPS pleomorphic adenoma has not been reported in the pediatric population. The tumor was removed using a combination of semi trans-parotid and trans-cervical approaches without mandibulotomy. Since the inferior limit of the tumor was at the inferior limit of the parotid gland, hence isolated transcervical approach would not have provided adequate exposure. Hence, semi trans-parotid approach helped in easy access to the superior limit of the lesion, which extended to the skull base. Differentiating a deep lobe tumor from a de novo minor salivary gland tumor can be challenging on pre operative radiological evaluation. A fat plane between the deep lobe and the tumor is suggestive of a de novo minor salivary gland tumor; also, visualization of a separate normal deep lobe from the tumor can be considered to be confirmatory for the same. The use of microscope intra operatively can not only provide the required magnification to differentiate these two entities and can potentially reduce the incidence of facial nerve injury [25, 26].

The patient was operated on 1 year back and is on regular follow-up until the writing of this manuscript. No evidence of residual or recurrence was seen. The wound remained healthy throughout with no morbidities and without any neurological deficit.

To conclude, we believe the trans-oral approach can be reasonable for small, less vascular tumors limited to the pre-styloid PPS. The transcervical approach remains the first choice for larger tumors, and this may be combined with transparotid or transoral with or without mandibulotomy. However, a tailored approach has to be structured for each individual depending on the nature and extent of the lesion in order to minimize complication.

Conclusion

- Such a large Pediatric de-novo PPS pleomorphic adenoma has not been reported to date.
- A semi- transparotid in combination with cervical approach for parapharyngeal space is also being reported for the first time.
- Semi- transparotid cervical approach is advantageous for huge parapharyngeal tumors in following ways:
- Helps in an easy and controlled approach to the superior extent of lesions extending till skull base
- Helps avoid mandibulotomy and morbidities associated with the same
- Minimizes the risk of facial nerve paresis, as with a trans-parotid approach
- The approach should be opted in cases where lesion does not extend below parotid and delineation of the lesion is difficult with isolated cervical approach

Patient's perspective

When my wife first noticed a swelling below our daughter's left ear, we thought it was nothing to be concerned about and would go in a while. For the initial 6 months, the swelling remained the same size. We routinely kept asking our child if she was having any pain in the swelling, but she was doing ok. She had no difficulty eating food and was breathing comfortably. She could attend school and play with her friends like she always used to do. However, as time passed, the swelling slowly started increasing. We then started to wonder what was causing this. One day while she was brushing her teeth, we also noticed a swelling inside her mouth. We panicked seeing this and immediately went to a nearby hospital. There, the doctor asked about the problems and checked the swelling. She advised us to go to an ENT specialist for this problem. On consulting the ENT doctor, we were told our child could have a tumor in her neck.

We were shocked to hear this. We thought tumors happen only to older adults who drink and smoke. However, we were assured by the doctor that some tests needed to be done to check the type of disease. We got MRI and FNAC done and reviewed with the reports. We were very anxious, waiting for the reports to come and spent sleepless nights thinking about the future of our beloved daughter. After seeing the reports, the doctor told us it needed to be operated on. We were explained that surgery is the best possible treatment and the risks

associated with the surgery. The doctors were warm and comforting, and we had complete faith in them. With God's grace, the surgery was successful. Our daughter had mild discomfort for the first few taken because the drain was eventually removed. She could speak and eat normally and was discharged after a week. After taking 1 week's rest at home, she started going back to her school, and our lives felt normal again. The swelling inside her mouth also disappeared. We go to the doctor routinely every few months for a check-up. I am forever thankful to the almighty and the doctors who helped us through this challenging time, and I am incredibly grateful for the care our daughter received.

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Authors' contributions
RS and RS collected the data. SP and RS prepared the manuscript. AB, MM and MP revised the manuscript. SP and RS did the final editing and submission. The author(s) read and approved the final manuscript.

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Availability of data and materials
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Declarations

Ethics approval and consent to participate
Taken. Ethical Committee Name: Institutional Ethical Committee AIIMS Rishikesh. Ethical Committee Reference Number: AIIMS/IEC/22/508

Consent for publication
Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient/parent/guardian/relative of the patient. A copy of the consent form is available for review by the Editor of this journal.

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

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