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A Case of Bilateral Anaesthesia Mumps after Cleft Palate Repair

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Abstract

Background: Anaesthesia mumps is a relatively unknown postoperative complication following general anaesthesia. It is an extremely rare complication, especially when it occurs bilaterally. It is usually a diagnosis of exclusion and has a benign prognosis. **Aim:** This case report will share our experience dealing with anaesthesia mumps. The condition arises in various patients undergoing various surgeries, making its pathogenesis confusing. By sharing our knowledge, clinicians can learn about the possibility, management methods, and prognosis. **Case Presentation:** A 2-year-old child presented for a routine cleft palate repair and subsequently developed bilateral acute post-operative parotitis. She was detained for a couple of extra days, managed conservatively, and the swellings subsequently resolved. **Conclusion:** Knowledge of the condition's possibility, early diagnosis, and prompt conservative management lead to the early resolution of symptoms.

Keywords

Parotitis, Acute Transient Swelling, Pneumoparotitis, Ischemic Sialadenitis, Cleft Palate, Trendelenburg Position, Pneumosialadenitis, Surgical Mumps

1. Introduction

Anesthesia mumps are rare, acute, transient, and usually self-limiting swelling of one or both parotid glands. It is an extremely rare post-operative complication following general anaesthesia. It has been reported across various surgical disciplines, including plastic surgery, obstetrics and gynaecology, otolaryngology, orthopaedic surgery, arthroscopic and general and neurosurgery. Munde first described

acute parotitis after surgery in 1878 [1].

Attas, in 1968, was the first to describe transient swelling of the salivary glands after general anaesthesia, which Reilly then followed up in 1970, who coined the term "anaesthesia mumps," describing three patients from a cohort of approximately 1500 patients who developed swelling of the parotid glands following administration of general anaesthesia [2] [3].

A case report of a 10-year-old boy who underwent exploratory laparotomy for a perforated appendix and subsequently developed anaesthesia mumps was reported by Reilly DJ [3].

Yonkers AJ *et al.* studied 11 cases seen at the University of Nebraska and Omaha Veterans Administration Hospital, looking at the aetiology treatment and prognosis. They isolated poor oral hygiene and dehydration as the main predisposing factors to anaesthesia mumps. They subsequently recommended proper hydration, correction of nutritional deficiencies, treatment of dental caries and oral infections, minimal use of belladonna drugs and minimal trauma during anaesthesia to help reduce the occurrence of anaesthesia mumps post-operatively [4]. The cases were mainly managed conservatively using warm moist packs, mouth irrigation, hydration and antibiotics, with a few needing irradiations of the gland or surgical incision and drainage [4].

In 2007, Liu F *et al.* encountered two patients who developed anaesthesia mumps after spine surgery under general anaesthesia, with one of them developing a swollen gland as early as 6 hours after surgery. Both patients were treated conservatively on non-steroidal anti-inflammatory drugs, and they made a full recovery [5].

Kiran S *et al.* reported on a case of a healthy 35-year-old woman who developed acute pansialadenopathy during routine thyroidectomy surgery, causing airway obstruction. They identified a possible cause to be endotracheal intubation [6].

Since then, various reports of anaesthesia mumps after a wide variety of surgical procedures have been published.

Acute, temporary parotid salivary gland swelling after general anaesthesia is uncommon and has been labelled as anaesthesia mumps, surgical mumps and post-operative parotitis. Multiple mechanisms have been proposed for the etiopathogenesis of this condition, but all remain unsatisfactory. Although it is benign and usually resolves after a few days without any medical therapy and with no sequelae, the attending health team should be aware of this complication. However, we present an incident in a two-year-old child who had general anaesthesia for a Bardach two-flap palatoplasty because of a Veau type III cleft palate and developed "anaesthesia mumps". This report will ensure that surgeons and anaesthetists know anaesthesia mumps as a possible complication following general anaesthesia and can be reassured by its relatively benign course and prognosis. Occasionally, respiratory distress requiring more serious interventions has been reported to occur. More literature is needed regarding its existence in the paediatric cleft palate population. Our case involved palatoplasty for a cleft palate, which can

occasionally have respiratory obstruction as a complication post-operatively. In these rare circumstances, knowledge of the possibility of anaesthesia mumps is critical as its occurrence in these patients could be potentially dangerous. Cases of anaesthesia mumps after cleft palate repair are currently not widely documented in the literature.

We encountered a child who was admitted for a routine repair of a cleft palate under general anaesthesia and developed bilateral parotid gland swelling 48 hours postoperatively, which resolved after a couple of days. It will be the first such case reported in Ghanaian literature.

2. Case Report

A 2-year-old, 15 kg, ASA 1 female toddler diagnosed with an isolated cleft of the secondary palate toddler underwent standard anaesthetic assessment before repair of an isolated cleft palate. No abnormal cheek swelling was present before surgery. Her childhood vaccinations were up to date, including her mumps vaccination. She was adequately hydrated before surgery.

Anaesthesia was inducted with Sevoflurane after pre-oxygenation. IV Propofol 20 mg was given, and muscle relaxation was achieved with 1.5 mg of IV Vecuronium. She was intubated orally with a size 3.5 mmID cuffed south-facing endotracheal tube. Anesthesia was maintained with an Isoflurane-oxygen-air mixture with a FiO_2 (fractional inspired oxygen concentration) of 40%.

The ventilator settings were pressure-controlled ventilation (PINSP 15 cmH_2O), RR 21 cycles per minute, and tidal volume 110 - 120 ml. IV cefuroxime 420 mg was given as antibiotic prophylaxis. IV Paracetamol 150 mg and IV Fentanyl 10 μg were given for analgesia. IV fluid therapy was 50 ml/hour of 1/5 Normal saline in 4.3% dextrose.

A bilateral supra zygomatic maxillary nerve block was given using 2 ml of 0.25% bupivacaine, and the palate was infiltrated with a mixture of 1:100,000 adrenaline solution and 1% lignocaine for hydro dissection of the palate and vasoconstriction to reduce bleeding.

The heart rate ranged between 110 and 130 bpm (beats per minute), and oxygen saturation remained 100% throughout the procedure.

The procedure was performed in a head-down supine position to visualise the palate better.

At the end of the procedure, muscle relaxation was reversed with IV Atropine and Neostigmine. She was extubated, fully awake, and transferred to the recovery ward. Anaesthesia lasted 1 hr and 20 minutes.

The child was stable postoperatively but, on a postoperative day two, was found to have painless bilateral cheek swellings. The father, who noticed his child's jaws were abnormally enlarged, confirmed this. The swellings were located in the anatomic region of the parotid glands, as illustrated in **Figure 1(b)**. The patient also had a temperature of 37.8 degrees centigrade.

Examination of the oral cavity did not reveal any calculi in the Stenson's duct

causing an obstruction. Also, there was no discharge coming out of the duct opening.

The resident radiologist on call performed an ultrasound scan, and the swellings were confirmed to originate bilaterally from the parotid glands. The left superficial parotid gland was diffusely enlarged with a homogenous pattern and measured 4.1 × 3.5 × 0.9 cm in size. The right superficial parotid gland was also diffusely enlarged, measuring 4.2 × 3.4 × 0.8 cm. The surrounding muscles and subcutaneous tissue were unremarkable. A full blood count was also performed, and the results are below.

White blood cell count—9.37 × 109/L (normal)

Neutrophil count (%)—34.6% (low)

Lymphocyte count (%)—57% (normal)

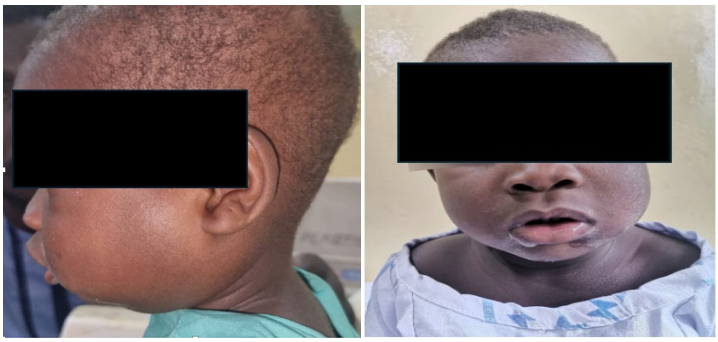
Eosinophil count (5)—0.4% (low)

Hemoglobin—8.1g/dl (low)

Platelet count—198 × 109/L (normal)

A diagnosis of post-surgical parotitis was made based on the bilateral diffused swellings of both parotid glands, which were not enlarged before surgery. The elevated temperature post-operatively also supported this diagnosis.

After conservative treatment, recovery was confirmed by the complete resolution of the parotid swellings and a normal temperature for 48 hours.



(a) (b)

Figure 1. (a and b) Photograph of the patient showing bilateral cheek swellings 48 hours postoperatively.

The parents were counselled and reassured about the new development.

Post-op management, as per our protocol, was IV cefuroxime for 24 hours and syrup paracetamol; good mouth hygiene, regular mouth washes, warm compresses and adequate hydration are beneficial. The swellings were resolved with no additional intervention.

3. Discussion

Acute post-operative parotitis was first observed by Munde, who in 1878 reported a case following ovariotomy [1]. Moricke, in 1880, recognised this complication in five out of his two hundred patients on whom ovariotomy was performed. He

concluded that it was specific for gynaecological operations and presumed a common neural "trigger path" between the gonadal and the parotid glands. Observing orchitis in epidemic parotitis and temporary parotid gland swelling during the menstrual cycle or amenorrhea underscored this theory [1].

In 1886, Paget modified Moricke's explanation to an "abdominal reflex theory" because most cases of people who had undergone an abdominal operation were recorded. Oehler postulated the haematogenous route of infection, but because of frequent bilateral occurrence, Wagner strongly opposed this view, suggesting a purely oral infection. Dyball, in 1904, expressed the belief that he had found the explanation for the direct damage to the parotid tissue in the action of bacterial toxins, mainly those of intestinal flora, which he presumed were released as a result of the abdominal or pelvic operation. The medical profession's interest in this subject is well demonstrated by Reischauer's 108-page article in 1931, which has more than 500 references [1].

Attas, in 1968, was the first to describe transient swelling of the salivary glands after general anaesthesia, which Reilly then followed up in 1970, who coined the term "anaesthesia mumps," describing three patients from a cohort of approximately 1500 patients who developed swelling of the parotid glands following administration of general anaesthesia [2] [3]. Yonkers described eleven cases of postoperative parotitis at the University of Nebraska and Omaha Veterans Administration Hospital from 1948 to 1971. These cases were studied from the standpoint of aetiology, treatment, and prognosis. The disease process was noted to occur in debilitated persons with poor oral hygiene, with symptom onset occurring from six to a hundred and seven days post-operatively but frequently within two weeks of surgery. Patients generally developed a red, swollen, tender parotid gland, leukocytosis, and a low-grade to moderate temperature elevation. The pathophysiology was postulated as retrograde staphylococcus aureus ascension through Stenson's ducts [4].

Since then, numerous additional case reports have been detailing similar findings. However, the cause and detailed pathophysiology of anaesthesia mumps remain unknown. Among the suggested causes in literature are trauma, head and neck positioning, straining and coughing during anaesthesia, vascular congestion, and venous engorgement of the head and neck. Overactive pharyngeal reflex stimulation of the salivary gland via the parasympathetic nerves, succinylcholine-stimulated copious secretions [5], dehydration, and mechanical blockage of the parotid duct by intubation and fixation of the endotracheal tube or head stripping and obstruction of glandular excretory ducts by position, calculi or thickened secretion were the major causes of acute salivary glands enlargement during induction of anaesthesia [6].

Morphine is known to reduce the secretion of amylase from isolated parotid acini. However, the ability of morphine to increase tone and decrease the propulsive activity of smooth muscle throughout the gastrointestinal tract is also well recognised. These latter effects may be mediated via μ receptors. The ducts of the

parotid salivary glands are surrounded by myoepithelial cells. These cells resemble smooth muscle cells, and by their contraction, they facilitate the movement of glandular secretions [7].

This condition is not limited to general anaesthesia. It has also been reported after regional anaesthesia due to dehydration or sympathetic stimulation caused by the perioperative use of vasopressors [8].

The incidence of Anaesthesia mumps, as reported in medical literature, is 0.2% - 17%, though the exact incidence remains unknown [3] [9] [10].

Anaesthesia mumps has a good prognosis and is self-limiting. Although most patients exhibit rapid resolution of the swelling between days one to five [8], some patients have required emergent intubation due to respiratory compromise [11]. Treatment is mainly supportive. Reassuring the patient and the family is a critical component of management, as well as close observation and rehydration. Where there is no contraindication, nonsteroidal anti-inflammatory drugs have been used to manage this condition [5] [12]. Positioning of the head and neck to allow normal venous blood circulation, especially when the patient is placed in a prone position or the duration of surgery is prolonged, is also recommended as part of measures to reduce the incidence. Minimum neck turning should be allowed to keep normal venous blood circulation, especially when the patient is placed in a prone position, or the duration of surgery is prolonged [13]. Premedication with anti-cholinergic drugs should be done to decrease secretions is also recommended [13].

Liu *et al.* [5], in their report of two cases following general anaesthesia (with an endotracheal tube), suggested that the presence of the patient's underlying disease (obesity), choice of anaesthetic drugs (succinylcholine, atropine), surgical position (prone, lateral decubitus), operative site (such as head and neck surgery) and induction methods (such as an endotracheal tube, laryngeal mask inadequate insertion and fixation) may all contribute to the development of acute swelling of the parotid glands after general anaesthesia.

From the review of existing literature, the only drug remotely linked to anaesthesia mumps used during the surgery for our patient was atropine [14] [15].

Even though various theories concerning aetiology have been proposed, the exact cause of post-anesthesia mumps remains unclear.

Some theories proposed include factors such as dehydration, neck hyperflexion during surgery, stenson duct obstruction, adverse drug reactions such as succinylcholine-induced hypersalivation, overactive parasympathetic tone, prolonged duration of surgery and venous congestion of the neck from trendelenburg positioning [16].

Cases of anaesthesia mumps have been published, and most of these predisposing factors were absent during the procedures. A patient who had a routine arthroscopic-assisted Hill Sachs repair for a recurrent shoulder injury under general anaesthesia was reported to have anaesthesia mumps post-operatively without any known significant predisposing factors other than general anaesthesia [17].

The differential diagnosis for acute post-operative head and neck swelling typically

includes jugular vein compression or thrombosis, venous congestion, hematoma, seroma, or chyloma. However, in the absence of a lymphovascular aetiology like the ones listed above," anaesthesia mumps" should be considered.

Other causes of parotid swelling worth considering and excluding include anatomical anomaly, viral or bacterial infection, prior radiation, sarcoidosis, amyloidosis, HIV infection, tuberculosis, Wegener's granulomatosis, Sjogren's syndrome, cystic fibrosis, and drugs such as iodides, phenylbutazone, thiouracil, L-asparaginase, and clozapine. The case presented had no features suggestive of these before surgery [18].

Although our patient had a short surgery and had face mask oxygenation, head down tilt and was administered atropine as part of her anaesthesia medications, it is unclear if they played a role in the development of her parotitis as these are standard procedures for all cleft palate patients in our unit. However, it is essential to consider these theories, especially in patients with a history of parotid disease or anaesthesia mumps.

Additional measures include minimising oropharyngeal stimulation, straining during intubation and extubation, and maintaining adequate hydration during the perioperative period [13]. Most cases are self-limiting and can resolve as early as 48 hours without intervention. In some more severe cases, re-intubation postoperatively may be needed. Also, in severe, recurrent infections, Stenson's duct ligation or partial Parotidectomy may be necessary [14].

4. Conclusion

In conclusion, this case report aims to draw the attention of surgeons and anaesthetists to this condition. Anesthesia mumps is a rare complication involving all age groups, various surgical procedures, and anaesthesia (Regional or general). Most cases are mild and resolve over a few days. Every anaesthetist and surgeon should be familiar with this uncommon complication and how it can be effectively managed.

Consent

Informed consent was obtained from the patient's parent to publish this case report.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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