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Case Report: Radiotherapy for the treatment of problematic drooling in a child with a life-limiting condition

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Abstract

Sialorrhoea or drooling can be a common problem in children with neurological conditions associated with bulbar dysfunction, possibly due to the overproduction of saliva or reduced swallowing of saliva. It is classified into anterior or posterior drooling.

There are many treatment options, from conservative management for anterior drooling such as oral/sensory motor therapy and behavioural intervention, to pharmacological and more invasive surgical intervention or botulinum toxin (Botox) injections for posterior drooling. Many interventions have been tried to achieve better saliva control, but there has been no consensus on the most effective treatment.

The use of targeted radiotherapy to the salivary gland to manage secretions in children is rarely documented but widely used in progressive neurological conditions in adults, such as Parkinson's disease and motor neuron disease.

In this case report, we review the use of targeted radiotherapy for managing drooling in a fifteen-year-old boy and the effectiveness of this treatment method.

Two years after treatment, the patient's drooling remains well controlled with one medication, and his quality of life has improved.

Keywords: Drooling, Radiotherapy, Paediatric palliative care.

Introduction

Sialorrhoea impacts around 40% of children with neurological impairment. However, those with a more severe cerebral palsy, with a Gross Motor Function Classification Score (GMFSC) III, IV, or V, are disproportionately negatively affected, at roughly 80% [1].

Search: MEDLINE, Embase, PubMed, CINAHL, and Google Scholar were searched using synonyms for "drooling" (sialorrhoea, hypersalivation, secretions), "radiotherapy" (radiation), and for "children" (paediatrics). No articles were found that discussed the use of radiotherapy in children to manage drooling.

Management of drooling is multi-faceted, involving members of the multi-disciplinary team, including, but not limited to, speech and language therapy, respiratory, surgical, physiotherapy, pharmacy, and neuro-disability teams.

Currently, there is no standardised treatment regime or protocol for managing oral secretions. Trials of various medications and a "wait and see" approach are often adapted to see what works for the individual. This often results in using multiple pharmaceutical agents to produce effective/acceptable saliva control.

The Drooling Impact Scale [2] measures the impact drooling has on the child and their family, assesses the impact of interventions over the short-, medium- and long-term, and documents the effectiveness of treatments.

McGeachan and McDermott suggest utilizing a four step treatment strategy – conservative management, anticholinergic drugs, Botox injections, and targeted radiotherapy [3]. However, it is noteworthy that they make this suggestion based on adults with neurological disease, as surgical intervention for drooling is rarely used amongst adults [4].

Radiotherapy is often seen as a treatment option after medical and surgical options have been exhausted [3]. In some instances, it may be prudent to discuss radiotherapy for patients who may be considered too high risk for a surgical procedure. However, a short anaesthetic for radiotherapy may be acceptable.

The risks associated with radiotherapy, particularly the potential long-term complications, must be considered carefully. There is a significant risk of over-drying the mouth, loss of sense of taste, and skin or mucosal irritation, plus the risk of radiation exposure in developing cancer at a later stage in life [5]. Palmer et al. discuss the risk of subsequent neoplasm after radiotherapy for childhood cancer, where they note that the higher the exposure and total dose, the higher the risk of developing a neoplasm. They found that solid tumours were the most common form of subsequent neoplasm, often occurring more than ten years after treatment [6]. Due to the life-limiting nature of some of the neurological conditions associated with severe drooling, the potential for developing neoplasm within this timeframe may not be a concern [3, 7].

Significant consideration must be given to the patient's age and the risk of musculoskeletal growth abnormalities, particularly for treating sialorrhoea, the risk of facial asymmetry, or disruption of teeth eruption [6].

As the results are often not permanent, additional treatments may be warranted. The risk of radiation toxicity and neoplasm increases with each treatment, and families should be counselled thoroughly about the risks involved.

Case Presentation

Patient A was a then 15-year-old boy with a medical history of cerebral palsy, GMFCS V, seizure disorder, scoliosis, recurrent chest infections, respiratory failure (non-invasive ventilation used for one year, not tolerated), and was gastrostomy fed.

He was known to multiple professionals in the tertiary hospital where he was managed. He had a history of frequent hospital admissions (3–4 per year) due to respiratory tract infections/aspiration pneumonia, particularly over the winter months.

A very social boy, he had a good quality of life, except for drooling, secondary to severe sialorrhoea. In March 2019, his mother reported to healthcare professionals that she was changing his bib a minimum of 15 times per day and using microfiber tea towels intermittently due to the excessive nature of his drooling.

He had a Drooling Impact Score estimated to be 77/100 (maximum scores for frequency, severity, bib use, wiping of mouth) and a Drooling Severity and Frequency score of 9/9.

He was treated with scopolamine patches (two one-point five mg patches per 72 hours, delivering 2 mg hyoscine) and glycopyrronium one mg three times daily. The addition of a trial of 1% atropine eye drops had no impact.

He had also previously been treated with an ultrasound-guided injection of botulinum toxin type A to the submandibular glands under the care of the Ear, Nose, and Throat team. He had botulinum toxin type A injections approximately six times for four winters; the last treatment was in November 2013. His mother reported that this was an effective treatment initially, but the effects wore off very quickly. Due to his deteriorating respiratory function, his parents felt the risk associated with the general anaesthetic versus the results from the Botox was not worth it.

Aside from the negative social impact drooling has, patient A was distressed by his drooling. This distress caused him to bite his lip, causing pain and injury to his lip (this further impacted his lip seal), which in turn increased his drooling and distress. The dental teams involved in his care had moulded gum shield/lip guard to prevent patient A from biting his lip, and while effective, these were poorly tolerated.

In June 2019, patient A was discussed and referred to a Radiation Oncologist for consideration for radiotherapy to address his hypersalivation. Risks associated with radiotherapy were explained, including xerostomia, mucositis, and the risk of later malignancy due to radiation exposure, in addition to the risks associated with general anaesthetic. His parents made the informed decision to go ahead based on the treatment's potential long-term positive impact. In October 2019, patient A had an 8Gy single fraction radiotherapy to his right-sided parotid salivary gland. Following treatment, he had mild mucositis, which was managed with regular oral care and paracetamol.

Six weeks after treatment, patient A was reviewed in the clinic at the tertiary hospital, and his anticholinergic medications were reduced to one point five mg scopolamine patch (reduction by 50%), and glycopyrronium reduced slightly to 1 mg in the morning and night and 0.5 mg in the afternoon with good saliva control.

Six months following treatment, his mother reports they had discontinued the scopolamine patch, and he was using glycopyrronium one mg three times daily, reducing slightly on warmer days.

Twelve months after treatment, his mother reported continuing on the single drug (glycopyrronium) and noted a significant improvement in the number of respiratory tract infections and the use of antibiotics in the preceding year. Patient A had no hospital admissions over the winter, which was unusual for him.

Patient A's Drooling Impact Scale had significantly reduced to 11/100 (the lowest score possible is 10/100), and his Drooling Severity and Frequency had reduced to 2/9 (the lowest score possible). He no longer bites his lip and no longer requires the gum shield.

Patient A continues to wear a bib, but this is used to dry his chin if he has stuck his tongue out. He no longer drools.

His mother reports that radiotherapy treatment has significantly impacted his quality of life, which was the aim of the treatment. Two years after treatment, his symptoms remain well controlled, and his medication use remains low compared to pre-radiotherapy.

Discussion

Sialorrhoea is uncommon in the general population of children over the age of 5 [8]. However, it is a common issue in children with neurological impairment, particularly those with cerebral palsy GMFSC III-V [1], thought to be related to bulbar dysfunction. Drooling can lead to chapped, macerated skin [3, 9]. Anterior drooling describes saliva escaping from the oral cavity, which may be considered a social problem. In contrast, posterior drooling can cause the pooling of saliva in the back of the throat, increasing the risk of aspiration.

There is a lack of good-quality evidence for any of the interventions currently offered. Additionally, the interventions for hypersalivation are non-curative and solely aimed at managing problematic and unpleasant symptoms [9]. A standardised measurement, such as the Drooling Impact Scale [2], to measure drooling before and after trialling an intervention is essential to evaluate the effectiveness of treatment and quality of life.

There are many different ways to manage excessive drooling, most of which are pharmacological, but surgical options are sometimes offered. There is a substantial risk of adverse effects

with pharmacological, botulinum toxin (Botox), surgical and radiotherapy options. The risk of potential side effects of each of these treatments should be explored prior to commencement. According to Khajuria et al., pharmacological options have the highest risk of side effects at up to 40%, with botulinum toxin A at 20% and surgical at 40% [9]. For radiotherapy, Hawkey et al. report short-term side effects up to 40% and 12% with delayed side effects [5].

Radiotherapy is not routinely or commonly offered to children with problematic drooling, even after the exhaustion of all other therapeutic options (non-pharmacological, pharmacological, and surgical). Hawkey et al. note that radiotherapy can profoundly impact the quality of life of those with neurological conditions [5].

Within adult neurology, in certain conditions such as Parkinson's disease and amyotrophic lateral sclerosis (ALS), radiotherapy to salivary glands is commonly considered when pharmacological options are no longer effective, or the side effects are troublesome [7]. A review by Shehee et al. compared radiotherapy and botulinum toxin in managing excessive drooling and found both methods effective, noting fewer significant adverse effects with radiotherapy [7]. However, they warn of the risk of neoplasm in later life as a potential side effect of radiotherapy – given the progressive nature of ALS, this may not be a concern due to the limited life expectancy with ALS.

In the cohort of children with severe neurological disorders, radiotherapy may be a suitable and relatively safe treatment option for excessive drooling as part of therapeutic options. Referral for suitability assessment should be considered and discussed with the child and their parents.

Summary

It is unclear whether salivary gland radiotherapy is not considered in children due to the potential risk of neoplasm or because it is not commonly recognised as a viable treatment option. Based on the effectiveness of treatment in this case study, children who are linked with palliative care or have a short life expectancy, and excessive drooling, should be considered for radiotherapy where there is a sub-optimal response to medications or botulinum toxin or when the side effect profile is challenging. A referral to a radiation oncologist for assessment should be made when there is a parental agreement.

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