

Paroxysmal Neuralgia in Pediatric Population- A Diagnostic Dilemma for Physicians and Dental Practitioners

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INTRODUCTION

Facial pain is a debilitating disorder if left untreated. It has been suggested that the most commonly undiagnosed facial pain conditions include neuropathic and myofascial pains because their pathophysiologies are not well understood. Facial neuralgias are otherwise rare in children. They are not acknowledged in most paediatric texts, there are few published reports on them.¹

The International Association for the Study of Pain defines neuropathic pain as "pain initiated or caused by a primary lesion or dysfunction in the nervous system".² Orofacial neuropathic pain is subdivided into episodic and continuous neuropathic pain. Episodic neuropathic pain, commonly called paroxysmal neuralgia, is characterized by episodes of sudden pain described as an "electric shock" usually lasting a few seconds and separated by refractory periods. Although rare, cases of trigeminal, glossopharyngeal, and occipital neuralgia present in pediatric patients.³

ABSTRACT

Paroxysmal neuralgia is relatively uncommon in children. Neuropathic orofacial pain is a challenge for the clinician, as no obvious dental pathology exists either clinically or radiographically. Dentist and physician should be able to recognize the characteristics of neuropathic pain so as to correctly diagnose these conditions hence avoid unnecessary dental intervention. This article reviews the conditions with paroxysmal neuralgia in children and available treatment strategies.

KEY WORDS

Glossopharyngeal neuralgia, neuropathic orofacial pain, trigeminal neuralgia

Epidemiology of paroxysmal neuralgia:

The age at which patients may be affected is more controversial. Adult neurological texts refer to idiopathic trigeminal neuralgia occurring occasionally in childhood and a number of children have undergone successful Microvascular decompression (MVD).^{4,5} However, there are few reports of Trigeminal neuralgia (TGN) occurring in pediatric literature.⁶

Trigeminal neuralgia is estimated to occur in 1 in 25,000 of the general population, with only 1% of these cases having an onset before the age of 20.¹ Lopes et al described two cases of trigeminal neuralgia in a six-year-old male and a 12-year-old female.⁷ Childs et al presented two more cases in 9- to 12-year-old male patients, while Manson et al described the youngest known patient with trigeminal neuralgia occurring in a 13-month-old.^{8,9} A case of TGN with

Chiari I malformation has also been reported.¹⁰ Matoth has reported a case of Idiopathic trigeminal sensory neuropathy in childhood.¹¹

Glossopharyngeal neuralgia (GN) is observed as a complication of tonsillectomy and in children with Chiari malformation type I.¹² Childs et al published a case of a 13-year-old female with glossopharyngeal neuralgia who presented with a history of right ear pain since infancy.¹⁰ The pain was initially attributed to otitis media without clinical evidence of inflammation. Kandt and Daniel described a 13-year-old male with a seven year history of GN triggered by stimulation of the right tonsillar area.¹³ Occipital neuralgia can occur in children with stenosis of the foramen magnum in achondroplasia and in cases of traumatic injuries, especially in adolescents.¹⁴

Clinical presentation:

The pain in TGN is paroxysmal, usually unilateral, and affects one or more divisions of the fifth cranial nerve. The most commonly affected single division is the mandibular branch, with the ophthalmic division the least often affected. The pain is often triggered by minimal stimulation of the affected area. Although pain may be experienced frequently throughout the day it characteristically does not occur at night.⁴

Glossopharyngeal neuralgia is characterised by bursts of severe, unilateral, lancinating pain of the same quality as that in TGN. Two forms seem to exist: one where the pain is mainly in the tonsillar area and throat, and the other where pain is felt deep in the ear. Mechanical stimulation of the fauces by swallowing, talking, and coughing typically triggers the pain.¹⁵

Specific features of pediatric neuralgia include scarcity of pain free episodes and an exaggerated debilitation leading to performance lack in essential activities.⁷ Glossopharyngeal neuralgia in the young has the potential to lead to developmental disorders, as the pain prevents the pediatric patient from eating.¹⁴

Etiopathogenesis:

Trigeminal neuralgia can be caused by tumours, vascular malformation, or multiple sclerosis. However, most cases are idiopathic and not associated with abnormalities on conventional magnetic resonance (MR) imaging.¹⁶ There is now convincing evidence that in these so called idiopathic cases pain results from vascular compression of the nerve root.⁸ Given that atherosclerosis is a physiologic process that starts in early childhood, vascular compression may be considered a cause of neuralgia pain in children.¹

Ecker and Smith proposed that reactivation of latent TGN is following reactivation of latent herpes simplex virus type 1. Surgical procedure of distraction osteogenesis in the affected side may also be an important predisposing factor for the recurrence in young population.^{17,18}

Koul R reported a case of TGN in a six year old girl which

was mainly caused due to arterial loop at the exit of TGN.¹⁹ Solth A reported a case of TGN in a 11 year old boy who had meningoencephalitis.²⁰ Trigeminal neuralgia has also been associated with lipomas in children.²¹

Glossopharyngeal neuralgia arises due to various causes such as tumor, infection, Chiari I malformation, infarction, dissection of the vertebral artery or neurovascular compression.²²

Childs et al have reported a case of GN in a 13 year old girl which was caused due to looping of the posterior inferior cerebellar artery.⁸

Evaluation/Diagnosis^{3,8,23}

The diagnosis of neuralgia is established by the history in combination with clinical examination and brain imaging studies. The clinical examination consists of both cranial nerve and a thorough intraoral assessment. A comprehensive intraoral examination of hard and soft tissues, including the tonsils and the oropharynx, is essential to rule out dental or oral infections as possible sources of neuralgic pain. If appropriate, an orthopantomogram and intraoral radiographs should be obtained to exclude dental and jaw pathology.

Once a clinical diagnosis of episodic neuropathic pain is established, prompt referral to a neurologist for comprehensive work-up is recommended. Assessment of pain and sensory testing in children is challenging. Validated indices of neuropathic pain in adults may be useful in children as well, but the developmental factors that are central to pain experience and expression in the young need to be considered.

Additionally, adolescents with chronic pain are more prone to impaired social functioning and other psychosocial issues, such as depression and anxiety. Referral to a clinical psychologist or psychiatrist may be warranted.

Diagnostic brain imaging studies should be performed in any pediatric patient with signs and symptoms consistent with paroxysmal neuralgia, as there is the likelihood of a serious underlying process such as multiple sclerosis or brain tumor. Magnetic resonance imaging (MRI) is preferred for the evaluation and diagnosis of paroxysmal neuralgia. MRIs demonstrate the anatomical relation between nerves and adjacent lesions.

Accurate and prompt diagnosis is significant in case of children. As neuralgia is typically characterized by unilateral pain, the differential diagnosis should include disorders that manifest as pain localized to one side of the head and face. Delayed diagnosis and consequently delayed institution of appropriate treatment may exacerbate the distress of both child and their family. Childs reported GN in a 13 year old girl who missed on average 12 days from school per term for years before the correct diagnosis was made. In her case, the diagnosis of recurrent otitis media was made with persistently normal examination findings on auroscopy.

Treatment:

One can either opt for surgical intervention or a more conservative therapy with pharmacotherapy. Before deciding on the course of treatment, an informed discussion with the child and parent(s) regarding the nature of the disorder and treatment options should take place.³

Pharmacotherapy:

Few controlled studies have been performed on interventions for neuropathic pain in children. Even the most commonly used first-line interventions, certain types of antidepressants and antiepileptic drugs, are almost exclusively prescribed on the basis of data from adults.²⁴ In 2006, Golden et al published a review of nonepileptic uses of antiepileptic drugs in the pediatric population and found no published trials evaluating the safety or efficacy of antiepileptic drugs in children.²⁵ Raieli et al administered carbamazepine (15 mg/ kg/day) to an 8-year-old patient suffering from trigeminal neuralgia with good results.²¹ A case of trigeminal neuralgia in a 12-year-old female was controlled using gabapentin (1,200 mg/day) in combination with carbamazepine (800 mg/day).⁷ Besides anticonvulsants, high dose courses of amitriptyline, phenytoin, baclofen, sodium valproate, sumatriptan, and acyclovir, together with a three day course of methylprednisolone have also been tried.⁸

Surgical treatment:

If medical treatment fails or the adverse drug effects are too pronounced, surgery may be required. Several surgical approaches are used for treatment of trigeminal neuralgia such as radiofrequency gangliolysis, glycerol gangliolysis, balloon compression, stereotactic radiosurgery, peripheral neurectomy, cryotherapy, and microvascular decompression.⁷

Resnick DK et al reported that patients whose symptoms begin in childhood do not enjoy the same therapeutic response to MVD as do patients with TGN onset in adulthood. An increased incidence of venous compression was noted in this population, as was a longer duration of symptoms before MVD. These factors may be responsible for the decreased efficacy of MVD in this patient population.¹⁶ Microvascular decompression is a more complex surgery to be performed in children than in adults because of the involvement of varying number of vessels. Children also seem to take a longer time to recover following the procedure. The most favored gamma knife surgery needs well-defined indications to perform in children considering the amount of radiation.¹⁸ However Solth et al have got good results with MVD using gore tex as tissue implant in a 11 year old boy.²⁰ Surgical decompression has found to be effective in children with GN.¹² Kandt RS performed cervical sectioning of glossopharyngeal nerve and its tympanic branch in a 13 year old boy and the child was completely relieved of symptoms.¹³

Percutaneous balloon compression has been performed by

Baabor MG . The low cost, low morbidity, low recurrence rate and high positive results make this procedure a valid option in the treatment of trigeminal neuralgia refractory to medical treatment.²⁶

Peripheral glycerol injection(PGI) has also been tried for the relief of facial neuralgia in children. PGI is less less formidable procedure, simple to perform and easily repeated so remains the choice for the majority of sick children with intractable TGN, along with the additional benefit of no risks of facial sensory loss when compared to that of classic neuroectomy.²⁷ Peripheral nerve stimulation serves as a good alternative to destructive surgical manipulation in occipital neuralgia.²⁸

DISCUSSION

Both glossopharyngeal and trigeminal neuralgia occur in children and can cause severe disability. These diagnoses should be considered in children presenting with paroxysmal facial, ear, or throat pain so that effective treatment can be given and investigation with MR angiography carried out.⁸

Paroxysmal neuralgias may cause symptoms that mimic dental pain and, consequently, lead to inappropriate treatments, including endodontic therapies or extractions of healthy teeth

Paroxysmal neuralgia may also be mistaken for jaw pain, as it can be triggered by jaw movements. For this reason, the pediatric dentist and physicians should know how to distinguish between paroxysmal pains triggered by mandible movements compared to musculoskeletal pain.³

CONCLUSION

The role of a pediatric dentist when treating a child diagnosed with neuropathic pain is to monitor for disease progression and to rule out any muscular or dental source of the pain. The pediatric dentist can collaborate with pediatrician and neurologist to both motivate and assess the treatment of a child who is suffering from neuropathic pain, thus directly improving the child's quality of life.³

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