Acta Scientific Otolaryngology (ISSN: 2582-5550)

Volume 5 Issue 4 April 2023

Case Report

Recurrent Bell's Palsy - A Case Report and Review of Literature

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DOI: 10.31080/ASOL.2023.05.0547

Received: January 11, 2023

Published: March 07, 2023

Abstract

Bell's palsy (Idiopathic Facial Nerve Palsy) is the most common cause of unilateral facial paralysis worldwide. However recurrent paralysis of the facial nerve is an uncommon occurrence and reported in only 7-8% of all Bell's palsy cases. Recurrent facial palsy was first reported in 1871. Recurrences of Bell's palsy in a patient may be the sign or symptom of a serious illness/syndrome. Therefore; in recurrent cases, family history, systemic diseases and malignancy must be investigated. We are reporting a case of recurrent bilateral Bell's palsy with two episodes within one year, with review of similar case reports/data available at present.

Keywords: Facial Paralysis; Idiopathic; Recurrent

Introduction

Idiopathic facial paralysis (Bell's palsy) is regarded as a benign common neurological disorder of unknown cause. It has an acute onset and is almost always a mononeuropathy of the seventh cranial nerve, and Dong SH., *et al*. [1] in their metaanalysis study has reported mean incidence of recurrent Bell's palsy as 6.5%. However Pitts DB., *et al*. [2] in their study of 140 patients with recurrent Bell's palsy, found recurrence in 2-9% of the cases. Swami H., *et al*. [3] has cited that the mean recurrence interval is usually more than one year in any case of recurrent Bell's palsy. In this case recurrence was reported within one year and patient recovered completely in both incidents, making it a unique case to report.

Case Report

A 42 year old lady presented with the history of sudden onset of weakness of the left half of face of four days duration. The weakness was associated with left side headache, with inability to close the left eye fully, moderate deviation of angle of mouth to the right and there was no drooling of liquids from the left side of the mouth. There were no wrinkles on left side of forehead. There was no history of traumatic injury to the face. There was no evidence to suggest Ramsay Hunt syndrome, and no plication of the tongue or labial oedema to account for MelkelssonRosenthal syndrome.

Patient gave history that she had similar complaints three months back, but the severity was more at that time. During both the incidents, there was no history of fever, vertigo, impairment of hearing or loss of taste. Similarly there was no history of exposure to cold or stress, prior to the episode of facial palsy preceding any of the two episodes. She was otherwise well and healthy with no known history of hypertension and diabetes mellitus. There was no other family member involved.

The patient had grade III facial nerve paralysis according to the House Brackmann facial nerve stages grading system. Rest of her ENT examination and remainder of her physical and neurological examination was normal. Her blood count, erythrocyte sedimentation rate, C-reactive protein, lipid profile, RA factor, thyroid function tests, ANA titre and vitamin B_{12} values were normal. As she had headache in the second incident, Her CT scan head was advised and it came out to be normal.

Patient She was treated with a course of oral prednisolone 1 mg/kg body weight daily on tapering dose for twenty days and oral acyclovir 400 mg five times daily for seven days. She was advised to use eye pad for eye protection during sleep and artificial tears were prescribed. She was also referred for facial physiotherapy with massage of the weakened muscles. At one month follow-up, the facial weakness had improved completely.

Discussion

Predisposing causes for recurrence of idiopathic facial palsy in an individual to are not well known, however Yurrita BS., *et al*. [4] have suggested its association with malignant hypertension, diabetes mellitus. Similarly in a study by Devriese., *et al*. [5] it was found that pregnancy, hypertension and diabetes mellitus increase the risk of developing recurrent idiopathic facial paralysis [6].

Katusic SK K., *et al*. in their retrospective study of patients with Bell's Palsy, proposed slight female preponderance in incidence. They further recorded in their observation that 86% of the patients whose record was evaluated had complete recovery. They identified complete facial weakness, non-ear pain, and hypertension as the most important risk factors for incomplete recovery. There study suggested that those having any of above risk factors and treated with steroids fared better than other treatment modalities.

Cirpaciu D., *et al*. [7] has conducted a retrospective study on a 10-years period for adults and a five-year period for children and reported that recurrent idiopathic facial palsy and Melkersson-Rosenthal syndrome was diagnosed more often in young females. They further observed that recurrence is more likely to occur in the first two years from the onset, so they suggested that patients diagnosed with Bell's palsy should have a follow up of at least two years from the onset. An interesting finding in their study was that the condition resolves completely in around 71% of the untreated cases. In this study, no significant difference between the affected sides of the face was found. The highest incidence of patients with Bell's palsy recurrences was identified between 21 an 30 years of age, and The lowest incidence between the ages 51 to 60.

Menassa J., *et al*. [8] has reported that recurrent idiopathic facial paralysis might occasionally be a symptom of the neurological involvement of Behçet's disease. Behcet disease is an auto-inflammatory systemic vasculitis of unknown etiology. It is characterized by recurrent oral and genital ulcerations, chronic relapsing uveitis, and systemic vasculitis involving arteries and veins of all sizes.

Kanerva M., *et al*. [9] has proposed Melkersson-Rosenthal syndrome as a rare cause of recurrent idiopathic facial paralysis. In Melkersson-Rosenthal syndrome, there are recurrent episodes of swelling of the lips, face and tongue along with recurrent facial paralysis attacks. The facial palsy in this syndrome can commence even in childhood and it may precede or coincide with the onset of edematous attacks. The edema usually affects the upper lip and cheeks [10]. Since there was no indication of these symptoms in our patient's history, these syndromes were not considered as a possible diagnosis.

Baloglu HH., *et al*. [11] has considered Lyme disease among the very common causes of bilateral or recurrent facial paralysis. This holds true in places where it is endemic like USA, Canada and middle east countries. In India it is not common. Lyme disease is characterised by erythema migraines, fever, fatigue or arthritis As patient did not have any such symptom or such history in past, the possibility of Lyme disease was excluded in the differential diagnosis. They have considered the possibility of Ramsay Hunt syndrome as cause of recurrent idiopathic facial paralysis while observing that it is very rare cause. They have quoted the study published by Pino Rivero V., *et al*. [18] to support their theory.

Multiple recurrences may present as part of other neurological disorder like Charcot-Marie-Tooth disease or as an autosomal dominant, variable penetrance hereditary Bells Palsy [12]. In their study to compare genetic predilection and recurrence tendency between facial palsy in Melkersson-Rosenthal syndrome (MRS) and Bell's palsy, Sun B., *et al*. [13] has found that facial palsy in MRS has an obvious genetic predilection and recurrence tendency as compared to Bell's palsy. They conclude this on the basis of data which showed that 31.3% of the patients with MRS have a familial history of facial palsy among their first or second-order relatives, but only 6.5% of the patients with Bell's palsy report familial history ($P < .01$), postulating that a genetic factor is strongly suggested in the etiology of facial palsy in MRS compared to Bell's palsy.

Recurrences on the same side require evaluation to rule out malignancy particularly schwannoma. Fibrous dysplasia of temporal bone has also been suggested as an etiologic factor for

recurrent facial palsy. An abnormally narrow facial canal may be a predisposing genetic factor as well.

Regarding the site of involvement of facial nerve, Fisch U., *et al*. [14] found that the edematous sites of facial nerve were proximal to geniculate ganglion in Bell's palsy during total facial nerve decompression, consistent with their results of intraoperative electrical stimulation, demonstrating that inflammatory sites of Bell's palsy were probably located proximally to geniculate ganglion.

Kurca E., *et al*. [15] has also propagated inflammatory pathogenesis Bell's palsy. They postulated that considering the fact that no one common infectious agent could be identified with certainty, the recurrent nature of Bell's palsy episodes was autoimmune in nature. The target attacked by the immune system was probably a neuronal antigen relatively specific for the intracanalicular facial nerve part as per their hypothesis. According to them an underlying autoimmune cascade, may be triggered spontaneously or by a multitude of factors including infection by cytomegalovirus/Herpes simplex virus or Epstein-Bar virus, insect bite, cold weather etc They have also mentioned in their study that two attacks are usually separated by more than 1 year and 70% of Bell's palsy relapses happen within 10 years following the original episode.

Pitts., *et al*. [2] in their study of 140 patients with recurrent facial palsy observed that recurrent facial palsy did not indicate a worse prognosis for recovery regardless of which side was affected. In this study, 77 patients were followed up for a mean of 33 years and they showed no progressive facial nerve dysfunction or tumor. They also postulated that younger the patient with Bell's palsy, higher is the probability of recurrence, and the probability of recurrence increases with the total episode count.

However, in a retrospective study Van Amstel., *et al*. [16], has proposed that the probability for the full functional recovery decreases with the total number of Bell's palsy recurrences. They further observed that in the case of ipsilateral recurrent paralysis, the response to treatment was found to be worse.

According to Kurca E., *et al*. [15], available evidence suggests that steroids are probably effective and acyclovir (combined with prednisone) is possibly effective in improving facial functional outcome. They also held that Rheologic drugs (e.g., pentoxiphylline) and vitamins (e.g. vitamin B_{12}) have uncertain therapeutic benefit though cyanocobalamin may possess a proneurotrophic effect. They observed that surgical approach may have its place in patients with incomplete recovery. They quoted that several authors have reported success with anastamosing affected facial nerve either with ipsilateral hypoglossal or accessory nerves or with the contralateral facial nerve using a surd nerve graft as a conduit.

Hato N., *et al*. [17] has observed that prednisolone along with antiviral like valacyclovir was more effective in treating Bell's palsy, than the conventional prednisolone therapy. This observation also supports the hypothesis that patients may be predisposed to facial nerve inflammation and demyelination secondary to exposure to an inciting antigen leading to cross-reaction with neuronal antigens specific to the intra-canalicular part of the facial nerve(Viral and auto immune theory of Bell's palsy).

Cirpaciu D., *et al*. [7] used hyperbaric oxygen therapy in their study for moderate to severe Bell's palsy, and suggested a benefit, but further added that there was not enough evidence to support this therapy. After analyzing the patient's records, they believed that those who suffered many episodes of facial palsy on one side, usually presented a complete facial paralysis and had a poor recovery. They didn't find any conclusive evidence to prove that Bell's palsy is a vasculopathy in vasa nervorum of the facial nerve, or has viral etiology. They negated the theory that Bell's palsy was associated with cold exposure, as it also appeared in the warm seasons, further another question was raised by them in their study: why is only one side affected when both left and right side are exposed. So they concluded that Bell's palsy remains a diagnosis by exclusion.

Teixeria LJ., *et al*. [19] in their study of evaluating various articles about various physical therapy strategies and devices, observed that facial exercises may reduce recovery time and decrease sequelae.

Image 1: Pt showing deviation of angle of mouth to right and loss of wrinkles on left side forehead.

Conclusion

In conclusion, it can be held that recurrent Bell's palsy is uncommon phenomenon and may have familial predisposition. Systemic diseases of diabetes and hypertension should be considered as an underlying reason in cases of recurrent idiopathic facial paralysis. Further, different etiological factors must be ruled out as a cause for such recurrence.

Conflict of Interest

No conflict of interest is declared by authors.

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