Irreversible facial nerve palsy as a revelator of parotid gland cancer

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Abstract

Aim of the study. The goal of the study was to assess the frequency of malignancies hidden under the diagnosis of "Bell's palsy". We aimed to create diagnostic algorithm to avoid failures concerning patients whose only symptom of parotid gland cancer was irreversible FNP. Materials and methods. We analyzed 253 consecutive patients with FNP treated in our department in the last 5 years. All patients with irreversible FNP were reassessed in 6-12 months. We underlined all shortcomings in the diagnostics of those in whom malignancies were found out in MRI of the neck and presented the proposal of diagnostic algorithm to avoid missing such an entity. Results. Bell's palsy was observed in 157 / 253 patients (62.06%), in 36 / 157 (22.92%) it remained permanent. In 4 / 36 patients (11.11%) with irreversible FNP, which constituted 2.54% of all "Bell's palsy" cases, parotid gland deep lobe mass was found out in MRI. In one patient infiltration of the skull base was diagnosed. Adenoid cystic carcinoma was confirmed in final histopathology in all cases. Conclusions. Our experience has shown that irreversible FNP can be a revelator of the malignant tumor located in the deep lobe of the parotid gland. Contrast-enhanced MRI covering intra- and extracranial segments of facial nerve should be ordered in all cases of FNP without recovery after 4 months. The main point of our study is to underline that the assessment of the deep lobe of the parotid gland with MRI should be included in the standard diagnostic protocol in all irreversible "Bell's palsy" cases. Key words: Bell's palsy, facial nerve palsy, parotid gland cancer

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Key points:

All patients with irreversible facial nerve palsy require further diagnostics to exclude organic, infectious, metabolic and autoimmunological cause of such a process.

Clinicians should pay special attention to patients with new or worsening neurologic findings at any point, ocular symptoms developing at any point, or incomplete facial recovery 3 months after initial symptom onset.

The differential diagnosis should put emphasis on expansive processes, including cancers.

Depending on the suspected localization of the lesion, high-resolution CT of the temporal bone and MRI with contrast enhancement are recommended for the evaluation of the facial nerve.

In selected undiagnosed cases, repeated imagings or even explorative surgery with biopsy of tissues adjacent to the facial nerve should be considered.

Introduction

Bell's palsy accounts for approximately 65% of acute facial palsies.[1] Diagnosis of Bell's palsy is still one of exclusion. Its etiology has been the subject of debates for years .[2-4] Nowadays, most authors refer to the theory of inflammatory demyelinating neuritis caused by reactivated latent viral infection (herpex simplex virus 1, herpes zoster virus) or other pathogens like Coxsackie virus, adenovirus or Epstein-Barr virus. Among non-infectious causes atherosclerosis and autoimmunological disorders (like Hashimoto or Sjogren disease) leading to facial nerve edema are listed.[3] Facial nerve palsy (FNP) has occasionally been reported as a complication following the administration of some vaccinations, including mRNA vaccines (BNT162b2 and mRNA-1273) against coronavirus disease 2019 (COVID-19).[5-6] Most researchers show that over 70% of patients with Bell's palsy make a full recovery in one week to six months.[2,4] In some groups, involving people over 60, patients with severe pain or pregnant at onset, with complete paralysis, suffering from diabetes and high blood pressure, the course of the disease may be more severe, even with cases of permanent FNP.[2,4]All patients with irreversible palsy need further diagnostics to exclude organic, infectious, metabolic and autoimmunological cause of such a process. The detailed evaluation should be based on in-depth history, clinical neurological and otolaryngological examination, laboratory tests and imagings to check for any viral (HIV, human herpesvirus 6, mumps virus, cytomegalovirus, and rubella virus), bacterial (Borrelia burgdorferi, Rickettsia, otogenic process), autoimmunological / metabolic (sarcoidosis, Sjogren's syndrome, sclerosis multiplex, GPA), and organic (tumors in the region of cerebellopontine angle, parotid gland, petrous bone, brainstem) pathologies.[2-4]

The goal of the study was to assess the frequency of malignancies hidden under the diagnosis of "Bell's palsy". We aimed to create diagnostic algorithm to avoid failures concerning patients whose only symptom of parotid gland cancer was irreversible FNP. One can use the algorithm for daily clinical practice and initiation of further studies.

Materials and methods

We analyzed 253 consecutive patients with FNP treated in our department in the last 5 years. The STROBE guidelines has been followed. The subject of the study were patients with Bell's paresis who were evaluated in detail. We reassessed all cases with permanent FNP and extended the diagnostics to exclude organic, infectious and autoimmunological cause of the disease. Contrast-enhanced magnetic resonance imaging (MRI) of the head and neck, laboratory tests for HIV infection, Lyme disease, autoimmune disorders including Sjogren's syndrome (if not available or already performed) were ordered. We describe four under-diagnosed patients with permanent FNP and malignancies found out in MRI and point out all deficiencies and shortcomings in their diagnostic process. Descriptive statistics was used to evaluate the study group. We also

present a proposal of diagnostic algorithm for patients with irreversible FNP.

The study was approved by an institutional ethics committee (KB-175/20). All patients provided written informed consent to participate in the study.

Results

Bell's palsy (idiopathic FNP) was observed in 157 out of all 253 patients with FNP (62.06%) who had reported to the outpatient department and emergency unit. Other reasons of FNP included: head trauma with skull base fracture, facial trauma, complication of acute or chronic ear infection, facial nerve schwannoma, middle ear squamous cell cancer (Table 1).

In Bell's palsy group 121 / 157 patients (77%) achieved a complete or partial regression of the face paralysis within 3 months, in 36 (22.92%) it remained permanent, despite typical treatment. In all patients with irreversible FNP, HIV infection and Lyme disease were excluded and the level of antinuclear antibodies was within normal limits. In 32 out of 36 cases the reason of permanent FNP was not found. In 4 patients (2.54% of all "Bell's cases" and 11.11% of irreversible "Bell's cases"), in MRI of the neck there was found a mass in the deep lobe of the parotid gland, in one patient infiltrating the skull base. The largest size of the tumor was 1.5, 2.0, 2.1 and 2.3 cm, respectively. Fine-needle aspiration biopsy confirmed malignant process while final histopathology adenoid cystic carcinoma in all analyzed cases.

The patients' details are presented in Table 2. In all cases the first symptom of parotid gland and skull base cancer was FNP, diagnosed as "Bell's paresis". In all patients, there was no palsy regression, therefore, further diagnostics was implemented. It included high resolution computed tomography (HRCT) of the temporal bone to exclude masked ear infection, ultrasound examination of the neck and salivary glands with linear probe and neurological consultation with head imaging (in 3 cases computed tomography (CT) while in 1 case both CT and MRI). When all results were within normal limits, diagnostic procedures were discontinued. In the above 4 cases, after 6 to 12 months of observation, reassessment with contrast-enhanced MRI of the head and neck was conducted. In all patients malignant process in the deep lobe of the parotid gland with infiltration of the main nerve trunk was revealed, in the absence of other symptoms and finally with a relatively low tumor mass volume. All patients were treated surgically - in 3 cases total parotidectomy with facial nerve resection was performed, while in patient with infiltration of the skull base lateral petrosectomy was necessary. All patients had adjuvant irradiation and are still under control. In 2 patients there are no signs of local, regional or distant dissemination of the disease. In other 2 metastases to the lungs have been detected in the 11^{th} and 28^{th}month of follow-up.

A proposal of diagnostic algorithm for patients with irreversible FNP was presented in Fig.1.

Discussion

Typically, Bell's palsy does not progress and it is rather unilateral and self-limiting. In fact, the majority of cases recover spontaneously within 3 weeks, even if untreated [7], or remit within 4-6 months, but nearly always remission is complete by 1 year.[8] In 2013 the specialists' panel made recommendations that diagnostic imaging for patients with new-onset Bell's palsy should not be performed routinely. However, clinicians should reassess or refer to a facial nerve specialist those Bell's palsy patients with (1) new or worsening neurologic findings at any point, (2) ocular symptoms developing at any point, or (3) incomplete facial recovery 3 months after initial symptom onset.[9]

The differential diagnosis of permanent FNP, with particular emphasis on expansive processes, including cancers, is mostly discussed in the literature basing on case reports.[10-12] We present a set of patients with irreversible FNP and under-diagnosed malignancies finally found out in the deep lobe of the parotid gland. All of them were primarily misdiagnosed with Bell's palsy. While detailed diagnostics implemented 3 to 8 months after FNP onset excluded masked ear infection and head pathology, as well as did not reveal pathology in ultrasound examination of the neck and laboratory tests, further examinations were discontinued.

The most common imaging techniques used in the assessment of salivary glands are ultrasound, CT and

MRI. Ultrasound is a quick and relatively inexpensive method that can accurately depict most salivary gland neoplasms.[13]However, it should be underlined that it provides limited visualization of the deep lobe of the parotid gland. Moreover, penetration of ultrasound can be hindered in case of high content of fat in the parenchyma.[14] CT is frequently used as a more precise tool, but, due to relatively poor soft-tissue contrast, sometimes the accurate assessment of the tumor extent and differentiation of its malignancy potential can be impossible.[13] Nowadays MRI is a gold standard in evaluation of parotid gland.[15]Our study shows that it is very important to include MRI in the diagnostics of all patients with irreversible FNP and, what important, even to repeat the imaging in uncertain / untypical clinical picture of the disease. MRI should cover all anatomical segments of facial nerve, both intra- and extratemporal. In 3 out of 4 presented patients with malignancies of the parotid gland, MRI of the head and neck was not included in the diagnostic algorithm. In one patient MRI scanning did not cover the region of parotid gland where the malignant tumor was located, which was an obvious shortcoming. Adenoid cystic carcinoma of the deep lobe of the parotid gland, which was finally confirmed in all presented cases, is a slow-growing but very aggressive tumor characterized, among others, by perineural spread.[16]It is estimated that FNP is an initial symptom in about 60% of cases.[17] Referring to our study group, early diagnosis with MRI of the neck could have improved oncological results in all of the analyzed patients, especially those with the distant spread of the disease.

Zimmerman et al [18] concluded that MRI is a useful tool for exclusion of otogenic and neoplastic processes with a sensitivity of 83% and 88%. Quesnel et al [19] indicate imaging of parotid gland, brain and temporal bone at 4 months for patients with irreversible idiopathic facial paralysis. They support that recommendation by the natural history of Bell's palsy, in which early signs of recovery are seen in the majority of cases by this time. We strongly agree with such an algorithm. In case of no lesion identified, they suggest to repeat the imaging in 3 months or even to consider explorative surgery and biopsy of tissues adjacent to the facial nerve. In our opinion, surgical procedures, due to the potential complications and difficulties, should be performed only in selected cases.

Although there are a few papers on magnetic resonance (MRI) protocols for the malignancies manifesting as facial palsy[20-22], they are universally suboptimal and thus may fail to reliably differentiate neoplastic from inflammatory process even when interpreted by experienced radiologists. Chhabda et al[23] underline that selected segments of the nerve are better evaluated on certain imaging modalities, so radiologists should try to combine clinical picture with specific facial nerve pathologies to tailor the imaging test to best answer the clinical question. Gupta el al [24] recommend contrast-enhanced MRI as the first imaging in cases when the palsy cannot be definitively localized. They indicate high-resolution temporal bone CT to evaluate the fallopian canal if the lesion can be localized in the region of the mastoid, tympanic, or labyrinthine segments of the facial nerve and contrast-enhanced MRI if the cisternal or intracanalicular segments of the facial nerve or the pontine nuclei should be assessed. In our opinion, such recommendations are very good. The intraparotid facial nerve is best evaluated by gadolinium-enhanced MRI, with T1-weighted gadolinium-enhanced images.[12] In the inner ear pathologies the combined non-contrast and postcontrast T1-weighted and T2-weighted sequences are mostly used.[24]

Our paper has strong points and some limitations. Our attempt to create diagnostic algorithm in irreversible Bell's palsy to avoid missing a parotid gland cancer is unique. The main limitation is a small sample size. Nevertheless, deep lobe adenoid cystic carcinoma is a rare disease and it is difficult to expand the study group. The algorithm can be used for clinical practice and initiation of further studies.

Conclusions

Neoplastic etiology should always be considered as a cause of irreversible FNP that originally seemed Bell's. Our experience shows that FNP can be a revelator of the malignant tumor located in the deep lobe of the parotid gland. Contrast-enhanced MRI covering intra- and extracranial segments of facial nerve should be ordered in all cases of FNP without recovery after 4 months. Repeated imaging should be considered in undiagnosed cases. The main point of our study is to underline that the assessment of the deep lobe of the parotid gland with MRI should be included in the standard diagnostic protocol in all irreversible "Bell's palsy" cases.

List of tables and figures

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