Artículo Original

Bilateral vocal fold paralysis: a retrospective study with novel and extremely rare causes of the condition

Paralisia bilateral das cordas vocais: um estudo retrospectivo com novas e raras causas desta condição

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Summary

Background: Vocal fold paralysis is the immobilization of the vocal fold due to an impaired function of the vagus or recurrent laryngeal nerve.

The aim of this study is to describe the most relevant aspects of the bilateral vocal fold paralysis (BVFP) in a series of 18 patients.

Material and Methods: A retrospective study regarding the 2005 to 2016 period was conducted, concerning symptoms, etiology and treatment. Obtained data were compared with medical literature.

Results: Eighteen cases were enrolled. The mean age was 60 ± 16 years and the main symptoms at presentation were upper airway dyspnea in 15 (83%) cases, dysphonia in 11 (61%), stridor in 4 (22%) and dysphagia in 3 (17%).

Surgical trauma was responsible for 11 (61%) cases: 7 after thyroidectomy, 3 after cardiac surgery and 1 after mandibular surgery. The remaining 7 cases causes were: Parkinson’s disease, multiple system atrophy, neoplastic meningitis due to t-cell acute lymphoblastic leukemia, breast cancer with pulmonary metastasis, laryngeal sarcoidosis, brainstem abscess and one with idiopathic cause. Twelve patients (67%) were treated surgically. Six (33%) patients did not receive surgical treatment. During the follow-up, all the patients were successfully decannulated.

Conclusion: The overall clinical features and treatment of BVFP were in accordance with the medical literature. Some of the presented cases represented extremely rare causes such as neoplastic meningitis due to leukemia and laryngeal sarcoidosis. Surgical treatment resulted in the resolution of the dyspnea, but with vocal quality deterioration.

Keywords

Vocal Cord Paralysis, Glottis, Paralysis, Sarcoidosis, Meningeal Carcinomatosis
Resumo

Introdução: A paralisia da corda vocal é a imobilização da corda vocal causada pela lesão do nervo vago ou do nervo laringeo recorrente. O objectivo deste estudo é descrever os aspectos mais relevantes de uma série de 18 casos com paralisia bilateral das cordas vocais.

Material e Métodos: Estudo retrospectivo englobando os anos de 2005 a 2016. Os dados referentes aos sintomas, etiologia e tratamento dos pacientes foram obtidos e comparados com a literatura médica.

Resultados: O estudo incidiu sobre 18 casos. A média de idades foi de 60 ± 16 anos e os principais sintomas iniciais foram: dispneia alta em 15 (83%) casos, disfonia em 11 (61%), estridor em 4 (22%) e disfagia em 3 (17%).

O trauma cirúrgico foi responsável por 11 (61%) casos: 7 após tireoidectomia, 3 após cirurgia cardíaca e 1 após cirurgia mandibular. As restantes causas (7 casos) foram devido a: doença de Parkinson, atrofia multissistémica, meningite neoplÁsica por leucemia linfoblÁstica aguda de células T, neoplasia primária da mama com metástase pulmonar, sarcoidose laríngea, abscesso do tronco cerebral e um de causa idiopática. Doze pacientes (67%) foram tratados cirurgicamente. Seis (33%) casos não receberam tratamento cirúrgico. Durante o seguimento, todos os pacientes foram descanculados com sucesso.

Conclusão: O quadro clínico e o tratamento da BVFP estavam de acordo com a literatura médica. Alguns dos casos apresentados eram relativos a causas extremamente raras, como meningite neoplÁsica por leucemia e sarcoidose laríngea. O tratamento cirúrgico resultou na resolução da dispneia, mas com deterioração da qualidade vocal.

Palavras chave

Paralisia da corda vocal, Glote, Paralisia, Sarcoidose, Carcinomatose Meningea
Introduction

Vocal fold immobility can result from structural or neurologic pathologies of the cricoarytenoid joint complex and can affect one or both vocal folds\(^1\). Vocal fold paralysis is the immobilization of the vocal fold due to an impaired function of the vagus or recurrent laryngeal nerve\(^2\). This is usually a consequence of an injury of the nerve fibers along their course\(^3\), anywhere from the brain cortex to the larynx\(^3,4\) and can be caused by central nervous system (CNS) or peripheral nervous system (PNS) pathologies.

The clinical presentation of vocal cord paralysis includes respiratory distress and phonatory and/or deglutition impairment\(^1\). The main causes of the paralysis are surgical trauma, mainly thyroidectomy, and malignancy\(^5\).

The surgical treatment of bilateral vocal fold paralysis (BVFP) remains an actual point of discussion. In most centers, cordotomy, with or without partial aritnoidectomy is the most selected procedure\(^1\).

The main objective of this study is to describe the most relevant aspects of the BVFP: etiology, symptomatology and treatment, in 18 patients, and compare the results with the data from medical literature. The secondary objective is to highlight some particularities of our series rarest cases.
Material and Methods

A retrospective study from the Voice Department of a tertiary health care institution was conducted. Medical records from 2005 to 2016 were selected. Inclusion criteria were: 1) age > 18 years; 2) bilateral vocal fold immobility documented by laryngoscopy; 3) sufficient clinical information. Data was extracted concerning symptoms, etiology and, when performed, surgical treatment and post-operative results.

An electronic search of literature using the PubMed and ClinicalKey databases was conducted on vagus nerve anatomy and on etiology, clinical aspects and surgical treatment of BVFP. The results of the series were compared with data obtained from the literature review.
Results

Series

Twenty two patients with PBCV were initially selected based on diagnosis, but 4 were excluded because they do not fulfill the inclusion criteria. A total of 18 cases were then enrolled. The mean age of the patients was 60 ± 16 years, minimum 28 years and maximum 88 years. Eleven patients (61%) were female.

The main symptoms at presentation were upper airway dyspnea in 15 (83%) cases, dysphonia in 11 (61%), stridor in 4 (22%) and dysphagia in 3 (17%).

Surgical trauma was responsible for 11 (61%) cases: 7 after thyroidectomy, 3 after cardiac surgery and 1 after mandibular surgery. The remaining 7 cases had different causes: Parkinson’s disease, multiple system atrophy, neoplastic meningitis due to t-cell acute lymphoblastic leukemia, breast cancer with pulmonary metastasis, laryngeal sarcoidosis, brainstem abscess and one with idiopathic cause. Data were detailed in Table 1 and topography of the lesions was represented in Figure 1.

Twelve patients (67%) were treated surgically: 8 needed emergent tracheotomy because of acute upper airway dyspnea; 6 were submitted to a posterior cordotomy assisted by CO2 LASER; and 1 patient had a combined approach for lateralization of the vocal folds.

The remaining 6 (33%) patients did not receive surgical treatment: 3 patients improved their condition after the treatment of the basal condition, 2 patients improved without specific treatment and 1 patient needed ventilatory support during sleep with a continuous positive airway pressure device.

The patients submitted to a surgical treatment improved the dyspnea but had a postoperative permanent decrease of vocal quality. During the follow-up, all the patients who had tracheostomy were successfully decannulated.
### Table 1—Patient characteristics and clinical presentation

<table>
<thead>
<tr>
<th>Case N°</th>
<th>Dysphonia</th>
<th>Dyspnea</th>
<th>Stridor</th>
<th>Dysphagia</th>
<th>Others</th>
<th>Cause</th>
<th>Treatment</th>
</tr>
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<tbody>
<tr>
<td>1</td>
<td>+</td>
<td>+</td>
<td></td>
<td></td>
<td></td>
<td>Total Thyroidectomy</td>
<td>Tracheostomy; Cordotomy;</td>
</tr>
<tr>
<td>2</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td></td>
<td></td>
<td>Laryngeal Sarcoidosis</td>
<td>Tracheostomy Main disease treatment</td>
</tr>
<tr>
<td>3</td>
<td></td>
<td>+</td>
<td>+</td>
<td></td>
<td></td>
<td>Total Thyroidectomy</td>
<td>Cordotomy</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td>+</td>
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<td></td>
<td></td>
<td>Total Thyroidectomy</td>
<td>Tracheostomy; Vocal fold lateralization</td>
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<tr>
<td>5</td>
<td></td>
<td>+</td>
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<td>Partial Thyroidectomy</td>
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<td>6</td>
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<td>Parkinson’s Disease</td>
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<tr>
<td>8</td>
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<td>Cardiac Surgery</td>
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<td>9</td>
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<td></td>
<td></td>
<td></td>
<td>Mandible Surgery</td>
<td>Tracheostomy</td>
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<tr>
<td>10</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td></td>
<td></td>
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<td>Tracheostomy</td>
</tr>
<tr>
<td>11</td>
<td></td>
<td>+</td>
<td></td>
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<td></td>
<td>Total Thyroidectomy</td>
<td>Tracheostomy; Cordotomy</td>
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<tr>
<td>12</td>
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<td></td>
<td></td>
<td></td>
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<td>T-ALL Neoplastic meningitis</td>
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<td>13</td>
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<td>14</td>
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<td>+</td>
<td>+</td>
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<td>Pulmonary metastasis of breast cancer</td>
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<tr>
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<td></td>
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<tr>
<td>18</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td></td>
<td></td>
<td>Cardiac Surgery because of aortic dissection</td>
<td>Main disease treatment</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>11 (61%)</td>
<td>15 (83%)</td>
<td>4 (22%)</td>
<td>3 (17%)</td>
<td>4 (22%)</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>
Vagus nerve anatomy

Vagus nerve fibers travel in 2 distinct anatomic systems, the central nervous system and the perypheral nervous system. The first/upper motor neuron is in precentral gyrus, whose fibers descend to the knee of the internal capsule, medial 1/5 of the crus cerebri (in the anterior part of mesencephalon) and in medulla oblongata. Inside the medulla oblongata, the lower neuron lies in the nucleus ambiguus. The fibers leave this nucleus and direct to the postero-lateral part of medulla oblongata, where they emerge to form the extra-axial vagus nerve, and travel through the anterior or nervous portion of the jugular foramen. After crossing this area, the fibers descend through the neck inside the carotid sheath, between the internal jugular nerve and the internal/common artery, achieving the supraclavicular region. Near the superior aperture of the thoracic cage, the right inferior laryngeal nerve (recurrent laryngeal nerve) passes below the right subclavian artery achieving the right tracheoesophageal sulcus. The left inferior laryngeal nerve enters in the mediastinum, travels below the aortic arch, and direct to the tracheoesophageal sulcus. Both inferior laryngeal nerves ascend between the esophagus and the trachea and enter in the larynx, providing motor innervation to the transverse arytenoid muscle, lateral cricoarytenoid muscle, posterior cricoarytenoid and thyroarytenoid muscle. Vagus nerve schematic pathway is displayed in the Figure 1.
Figure 1- Schematic representation of the vagus nerve and of the level of injury by pathology. The arrows point to the most probable place of injury. A- Gyrus precentral; B- Internal capsule; C- Brainstem; D- Neck and thorax. Legend: aa- aortic arch; b- cranial base; c- left common carotid; cer- cerebellum; cX- central vagus nerve; h- hyoid; k- internal capsule knee; l- laryngeal motor cortex; lil- left inferior laryngeal nerve; m- mesencephalon; mo- medulla oblongata; na- nucleus ambiguous; p- pallidum; po- pons; pX- peripheral vagus nerve; ril- right inferior laryngeal nerve; sa- supraclavicular artery; t- thalamus; tir- thyroid cartilage; tr- trachea. Image based in 6, 8, 9. Image is not at scale.
Discussion

The mean age of the disease of our patients at presentation was 60 ± 16 years, agreeing with the literature: the mean age of BVCP at presentation is in the 5th to 6th decade of life, with a minimum of 18 years and maximum of 90 years.

Clinical manifestations were also in accordance with the literature, with dyspnea and dysphonia as the most prevalent symptoms. Stridor and dysphagia were also described at presentation.

As in medical literature, most cases of our series were due to surgical trauma, mainly thyroid surgery. The mechanism of the nerve injury identified in other studies includes neural disruption (caused by thermal damage, stretch, cutting or compression) or vascular compromise. The remaining post-surgical cases were due to cardiac procedures and mandibular surgery. According to some authors, BVFP due to cardiac surgery is more common nowadays, mainly because of an increasing numbers of heart and aorta artery procedures/surgeries.

The non-surgical etiologies of our series represented 39% of the cases. Two patients (11%) presented a neurologic etiology: Parkinson’s disease and multiple system atrophy. In other series, neurological causes are responsible for about 5% of all BVFP. The 2 neoplastic causes of our series (11%) were all extra laryngeal: leukemia (neoplastic meningitis) and breast cancer with pulmonary metastasis. In some series this etiology is responsible for 14% of all cases. In the literature review, only one case of neoplastic meningitis caused by hematogenous cancer was identified, a diffuse large-B cell lymphoma, so it seems that other cases of leukemic meningitis were not previous described in the English published literature.

The other two cases were caused by inflammatory or infectious disease: laryngeal sarcoidosis and brainstem abscess. After an exhaustive review on English literature we found only 5 cases of BVFP caused by laryngeal sarcoidosis. The last patient (5.5%) of our series had an idiopathic etiology. This etiology was responsible for 11% in other series.

With regard to surgical treatment, the main handicap of cordotomies, that is the most frequent procedure in our and other series, was the compromise of voice quality in spite of the excellent results on glottis patency, avoiding tracheotomy during the follow up. Cordotomy has advantages: simplicity, brevity, good results, few days of hospitalization, reduced risk of complications and is easy to perform a second surgery if needed.

In the last years, experimental techniques, like laryngeal reinnervation, had been developed in order to regain some of the vocal fold mobility and tonus. Reinnervation showed improvement of the muscular tonus, of the muscular atrophy and some improvement in the voice quality. In BVFP, the need of regain vocal fold mobility is paramount in order to decannulate the patients. In some specialized centers, reinnervation can be made using phrenic nerve and phrenic nerve root, with good results.

Some other developments had been explored, such as laryngeal pacers, with promising results.
Conclusion

The overall presentation and treatment of BVFP in our series was in accordance with the data of medical literature. Some of the presented cases represent extremely rare causes of bilateral paralysis of the vocal folds, such as neoplastic meningitis due to leukemia and laryngeal sarcoidosis. Surgical treatment resulted in the alleviation or cure of the dyspnea, but with deterioration of the vocal quality. Because this scientific field is currently developing, it is foreseeable that new and more effective treatments appear in the next years.

Conflict of Interest

The authors declare that they have no conflict of interest.
References


