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CASE STUDY

DIAGNOSTIC AND THERAPEUTIC APPROACH IN UNILATERAL DIAPHRAGMATIC PARALYSIS

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ABSTRACT

Unilateral diaphragmatic paralysis is a condition that can lead to respiratory dysfunction and, in extreme cases, death. Presents similar etiology to paralysis of bilateral involvement, and in many cases, are idiopathic. Therefore, have considerable diagnostic not index. Has wide range of clinical manifestations, ranging from asymptomatic episodes' frames with respiratory failure. This article makes a bibliographical review about the main features of unilateral diaphragmatic paralysis (UDP), highlighting the most relevant data about definition, etiology, pathophysiology, clinical presentation, diagnosis and more effective treatment of this illness to systematize the knowledge about this disease.

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INTRODUCTION

The diaphragm is the main muscle of inspiration and the most important of the respiratory muscles. Also, serves as mechanical barrier between the thoracic and abdominal cavities, besides keeping the difference in pressure between them. Each diaphragmatic hemi cupula is innervated by the phrenic nerve diaphragm ipsilateral (Stojkovic *et al*., 2004; MacBruce *et al*., 2016). Diaphragmatic paralysis (DP) may involve the entire diaphragm (bilateral) or just a brochure (unilateral), which is the most common form (MacBruce *et al*., 2016; Onders *et al*., 2014; McCool and Tzelepis, 2012). It is important to note that depending on the degree of commitment to new regions will have the function to take the job, the example of accessory muscles or diaphragmatic lesion contralateral portion (MacBruce *et al*., 2016). Soon, the paralysis of such muscle, uni or bilateral basis, is, in most cases, compatible with life due to the effective adaptation of the other muscles of respiration (Issahar Ben-Dov, 2012).

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Unilateral diaphragmatic paralysis (UDP) is a condition that can lead to respiratory dysfunction and features like the etiology of bilateral involvement. Patients are usually asymptomatic at rest, but can have a varied symptomatology. The diagnosis is usually suggested by typical findings on chest x-ray and confirmed by fluoroscopic assessment (MacBruce *et al*., 2016; León-Atance *et al*., 2011; Miki Oike *et al*., 2012). In symptomatic patients, is performed many times a resection of the diaphragm to prevent your paradoxical movement in inspiration. This time-honored approach brings relief to many patients, but the mechanism for this is not well understood. In addition, it is not known whether symptomatic relief also translates into an improvement in exercise tolerance (Freeman *et al*., 2009; Higgs *et al*., 2002). Knowing the variability of the presentation of the UDP and the high level of clinical suspicion for early diagnosis, the present work aims to bring together the most important and current studies on the topic.

MATERIALS AND METHODS

The present study is a review of the literature in PubMed/Medline, Scopus and Web of Science databases using the descriptors: "*respiratory paralysis", "muscle", "muscle*

paralysis", "diaphragmatic paralysis", "respiratory insufficiency", being evaluated cohort studies, literature review and case reports. We included English, Spanish and Portuguese language articles, published between 1997 and 2017, which portrayed the unilateral diaphragmatic paralysis. During the initial research, 139 articles were selected which have been assessed by the authors in accordance with the following inclusion criteria: articles published in Portuguese, English or Spanish, to provide the keywords in the title or abstract, published between the year 1997 and 2017. After the initial check, all abstracts and methods were read and those who have not covered the topic proposed, or that were repeated, were excluded. The final featured material 65 articles.

RESULTS

Pathophysiology

The diaphragm is a structure consisting of a central tendon surrounded by muscle fibers, being relatively symmetrical, becoming the most important muscle of breathing process (Issahar Ben-Dov, 2012). Thus, serves as a mechanical barrier between the thoracic and abdominal cavities, while retaining a pressure difference between them (MacBruce *et al*., 2016; Issahar Ben-Dov, 2012). Each diaphragmatic hemi cupula is innervated by ipsilateral phrenic nerve, which is a branch of cervical plexus, composed of fibers of C3 to C5 (cervical plexus). Already your outer border is innervated from the segments T7 to T12 (Thoracic plexus). The vascular supply comes phrenic arteries and the pericardiophrenic arteries (MacBruce *et al*., 2016; Miki Oike *et al*., 2012; Maish, 2010; Naunheim, 1998). The muscle fibers are inserted into the sternum, ribs and arched ligaments. Such conformation allows the contraction of fibers facilitates the elevation and expansion of the rib cage, expanding the senses and circumferential flow, so that the influx of air into the lungs (Issahar Ben-Dov, 2012; León-Atance *et al*., 2011; Miki Oike *et al*., 2012; Freeman *et al*., 2009; Higgs *et al*., 2002; Maish, 2010; Naunheim, 1998). It's worth pointing out that from the time of the paralysis, the typical flow offset this musculature (from a process of contraction) is severely reduced or abolished (Canbaz *et al*., 2004; Kaufman *et al*., 2014; Sacher *et al*., 2006). This scenario raises a pleural pressure more negative with inspiration, so that the offset is directed in cephalic sense. Because of this, there is a commitment of chest expansion and pulmonary ventilation, leading to a substantial reduction of tissue perfusion and hypoxia (McCool and Tzelepis, 2012; Singer *et al*., 2008; Verin
et al., 2006; Ansari *et al*., 2010).

Etiology

The UDP etiological aspects like features of bilateral involvement (MacBruce *et al*., 2016; Onders *et al*., 2014; McCool and Tzelepis, 2012; Issahar Ben-Dov, 2012; León-Atance *et al*., 2011; Miki Oike *et al*., 2012). Among them include idiopathic causes (responsible for up to 2/3 of cases) (León-Atance *et al*., 2011). Also, the following factors:

- Phrenic nerve injury during heart surgery, chest, liver transplant or cervical surgeries (Issahar Ben-Dov, 2012; León-Atance *et al*., 2011; Canbaz *et al*., 2004; Kaufman *et al*., 2014);
- Complication of cardiac ablation catheter (Sacher *et al*., 2006);
- Graduate electrical cardioversion because of phrenic nerve injury by the slight increase of temperature, or damage to the right internal mammary artery leading to the heart attack of phrenic nerve (McCool and Tzelepis, 2012); embolic phenomena that affect the function of the phrenic nerve (Singer *et al*., 2008);
- Malignant neoplasm and neurodegenerative diseases (Verin *et al*., 2006);
- Herpes zoster, polio, dengue and other diseases viral (León-Atance *et al*., 2011; Ansari *et al*., 2010);
- Cervical spondylosis and cervical compression tumors (León-Atance *et al*., 2011);
- Blunt trauma and neck surgery (León-Atance *et al*., 2011; Verin *et al*., 2006);
- Pneumonia and iatrogenic embolization (León-Atance *et al*., 2011; Chapman *et al*., 2000; McCaul and Hislop, 2001);
- An inflammatory disorder of the brachial plexus and brachial plexopathy hereditary (Issahar Ben-Dov, 2012; León-Atance *et al*., 2011).

Clinical manifestations

During anamnesis, the medical professional should research data about prior pathologies and elucidate the entire history of surgery, so that it is possible to indicate the causal factors of clinical manifestations (Chapman *et al*., 2000; McCaul and Hislop, 2001; Declerck *et al*., 2013; Hart *et al*., 2002). Patients with this pathology can experience a variety of symptoms (Perez, 2006; Jubran, 2006; Steier *et al*., 2008), which depend on the pre-existing cardiopulmonary condition, the extent/severity of paralysis, as well as the nature of the frame (acute or chronic) (Issahar Ben-Dov, 2012). Generally, the affected are asymptomatic at rest, but may present Dyspnea and respiratory performance reduction during exercise (Declerck
et al., 2013; Hart *et al*., 2002). In the presence of an underlying lung disease or intercurrent, there may be dyspnea during the home. Orthopnea, despite already being observed in some situations, it's not as intense as in diaphragmatic paralysis bilateral (Chetta *et al*., 2005; Abad *et al*., 2001; Podgaetz *et al*., 2016). It is important to note that this disorder may be intrinsically associated with respiratory sleep disorders (Le Pimpec-Barthes *et al*., 2014; Shields, 2005; Kuniyoshi *et al*., 2004). In addition, in its more serious presentations, the clinical picture can develop into respiratory failure and death (Freeman *et al*., 2009; Hüttl *et al*., 2004; Groth and Andrade, 2010; Groth *et al*., 2010).

Diagnosis

The diagnosis of unilateral diaphragmatic paralysis is usually suggested by typical findings on chest x-ray, seen in an upright position and confirmed by fluoroscopic evaluation (Issahar Ben-Dov, 2012; León-Atance *et al*., 2011; Mouroux *et al*., 2005; Freeman *et al*., 2006; Kara *et al*., 2015; Mouroux *et al*., 1997). In radiography there is a high asymmetric diaphragmatic, but that is not specific to UDP, being common to association with atelectasis above the paralyzed hemicúpula (Issahar Ben-Dov, 2012), (León-Atance *et al*., 2011; Naunheim, 1998; Chetta *et al*., 2005).

About the assessment, fluoroscopic examination can reveal a paradoxical rise during inspiration compared to the affected hemidiaphragm, i.e. during inspiration the muscle doesn't come down properly. This diverges from what is found on the

healthy side, that is, a quick descent of the hemidiaphragm normal. This test is positive in more than 90% of patients (Kim *et al*., 2007; Freeman *et al*., 2009; Hwang *et al*., 2003; Moon *et al*., 2000).

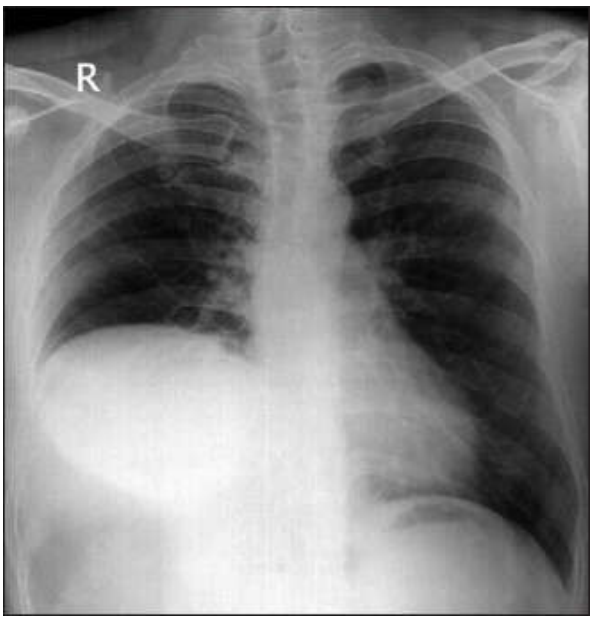

Figure 1. Chest radiograph showing elevation of the right diaphragmatic dome. Source: Ansari M K, Jha S, Nath A. Unilateral diaphragmatic paralysis following dengue infection. Neurol. India 2010; 58: 596-8

Figure 2. (A) Fluoroscopy demonstrating insignificant decrease of hemidiaphragm affected during inspiration. (B) Expiratory film showing insignificant rise of right hemidiaphragm, and no paradoxical movement. Source: Ansari M K, Jha S, Nath A. Unilateral diaphragmatic paralysis following dengue infection. Neurol. India 2010; 58: 596-8

The pulmonary function test also provides important data such as the degree of respiratory impairment. Thereby, the spirometry (supine and sitting positions) reveals, in the UDP, a forced vital capacity typically diminished (70% to 80% of predicted values) (Groth *et al*., 2010; Groth and Andrade, 2010). Another test of fundamental image is computed tomography of the chest, which is performed to evaluate the course of ipsilateral phrenic nerve and delete tumor involvement (León-Atance *et al*., 2011; Naunheim, 1998). Polysomnography can also be obtained to make possible pathological frame associations with sleep disorders (León-Atance *et al*., 2011; Celik *et al*., 2010; Simansky *et al*., 2002; Graham *et al*., 1997). However, the electromyography and transdiaphragmatic pressure measurement, have limited role in UDP (Higgs *et al*., 2002). In addition, the laboratory tests are nonspecific in most occasions (León-Atance *et al*., 2011; Freeman *et al*., 2006).

Differential diagnosis

The differential diagnosis of UDP includes pleural effusion supradiaphragmatic, diaphragmatic eventration, diaphragmatic hernia and unilateral and subdiaphragmatic processes.

Treatment

Most patients with UDP are asymptomatic and do not require treatment. The episodes with mild symptoms, usually improve spontaneously over the course of one to two years (Podgaetz *et al*., 2016; Ribet and Linder, 2002; Wright *et al*., 1998; Versteegh *et al*., 2007). Morphofunctional studies of neuromuscular respiratory chain chest and tests allow the selection of patients who are candidates for surgery. The surgical treatment is proposed for the patients with diaphragmatic dysfunction permanent and irreversible (Le Pimpec-Barthes *et al*., 2014; Graham *et al*., 1999; Leo *et al*., 2010; Leo *et al*., 2004). However, phrenic nerve conduction studies and electromyography are not widely available, so that the degree of phrenic dysfunction is usually unknown (Le Pimpec-Barthes *et al*., 2014; Graham *et al*., 1997). The aim of surgical treatment is to place the diaphragm paralyzed in a position of maximum inspiration that allows the compression relief about the pulmonary parenchyma, allowing your re-expansion (Shields *et al*., 2005; Versteegh *et al*., 2007; Hwang *et al*., 2003; Freeman *et al*., 2009). Surgical treatment can also be considered when the dyspnea is disproportionate to the degree of physical activity or to the severity of lung disease (Le Pimpec-Barthes *et al*., 2014; Higgs *et al*., 2002). The surgical approach of the hemidiaphragm affected has provided excellent results if well indicated (Podgaetz *et al*., 2016; Kuniyoshi *et al*., 2004; Freeman *et al*., 2009). Is performed using the open technique, thoracoscopy or laparoscopic, and involves creating and fluted suture in diaphragmatic muscle to reduce the mobility of the injured region (Podgaetz *et al*., 2016; Hüttl *et al*., 2004; Groth and Andrade, 2010; Groth *et al*., 2010). The techniques for thoracoscopy recently are being applied more (Mouroux *et al*., 2005; Freeman *et al*., 2006; Kara *et al*., 2015; Mouroux *et al*., 1997; Kim *et al*., 2007; Freeman *et al*., 2009; Hwang *et al*., 2003; Moon *et al*., 2000), and good results have also been reported with laparoscopic techniques (Groth *et al*., 2010; Groth and Andrade, 2010; Higgs *et al*., 2002; Welvaart *et al*., 2013; Celik *et al*., 2010). Open transthoracic approach include various dimensions, thoracotomy in various spaces intercostal, as conventional posterior and lateral side , but also sternotomy and the hemi-shell-shaped incision (Graham *et al*., 1997; Higgs *et al*., 2002; Freeman *et al*., 2006; Ribet and Linder, 2002; Wright *et al*., 1998; Versteegh *et al*., 2007; Graham *et al*., 1999; Leo *et al*., 2010; Ciccolella *et al*., 1999). As a result, reduction of dyspnea, improvement in pulmonary function evaluation on blood gas and resistance exercises (Podgaetz *et al*., 2016; Groth *et al*., 2010; Leo *et al*., 2010).

According to Leo *et al*. the suture line is run from the middle line of the dome of the diaphragm, the phrenic nerve level (Leo *et al*., 2004). Important to evaluate the extent of resection. Some authors defend the plication continues until the diaphragm is tense as possible or obvious on palpation (Graham *et al*., 1997; Versteegh *et al*., 2007; Hwang *et al*., 2003). Leo *et al*. stressed the importance of achieving a flattened Dome, allowing the diaphragm to assume normal curvature in the costal and diaphragmatic position (Leo *et al*., 2004). The studies of Freeman *et al*. and Higgs *et al*. showed that after resection of the diaphragm, spirometry and static lung volumes returned to within normal limits, but the changes in pulmonary function tests were relatively small and in patients undergoing a course plicature of the diaphragm, current volumes increased and decreased respiratory frequency (Freeman *et al*., 2009; Higgs *et al*., 2002). Despite this improvement, the exercise capacity remained unchanged, still become the most likely explanations for the symptomatic relief (Welvaart *et al*., 2013). In some cases, there may be need for non-invasive ventilatory support with positive mechanical pressure ventilation or invasive. The requirement for such ventilation methods are observed in cases where there is acute respiratory failure due to some infectious process, respiratory exacerbation of a pre-existing pulmonary pathology or after general anesthesia for surgical procedures (Qureshi, 2009). Typically, patients with UDP have an excellent prognosis, unless the underlying cause is associated with a severe and fatal pathology. However, the prognosis for recovery of diaphragmatic function is variable (Issahar Ben-Dov, 2012; León-Atance *et al*., 2011; Perez, 2006; Ciccolella *et al*., 1999).

In the long run, patients with diaphragm paralysis present objective and subjective improvement after resection, ensuring the remission of symptoms and improves quality of life (Celik *et al*., 2010).

Conclusion

No study is completely diagnosis to UDP, so that a high degree of suspicion is always necessary. The need for more tests varies according to the clinical situation. Most of the time, the discovery is at random, from a chest x-ray. Although most of the patients being asymptomatic or oligosymptomatic, as well as attend with an excellent prognosis without treatment, a select group of patients presents a more serious condition, requiring surgical treatment.

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