Letter to the Editor

Small pleural holes of the apical parietal pleura: an unknown precursors of cervical lung hernia and association with primary spontaneous pneumothorax

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Parietal pleura is the outer layer of the pleura, a serous membrane which covers the chest wall, the mediastinum, and the diaphragm (1-3). Anatomically, the apical pleural contains some "stomas" that are microscopic channels which allow the passage of the pleural fluid into and from the pleural space.

Anomalies of the parietal pleura are very rare and have rarely been reported as present at the apical parietal pleura (4-7). Some authors have described the presence of some defects (single or multiple, varying in size from a pinhole up to a centimeter or more in diameter) at the diaphragmatic pleural level responsible for pleuroperitoneal communications (8-12). These diaphragmatic pleural anomalies mainly occur at the tendinous portion of the central diaphragm and are common in the right hemidiaphragm (13). These defects are speculated to be induced by the following factors: congenital or traumatic diaphragm defects, defects or laceration of blebs in the fragile region of the diaphragm, and abnormal intraperitoneal pressure. In some cases, diaphragmatic pleural defects (holes, perforations, fenestrations, pores, porosities, stomata) have been associated with catamenial pneumothorax (14,15) or hydrothorax (11,12) and, in the majority of cases, they were surgically repaired by video-thoracoscopic surgery.

Defects in the apical parietal pleural are rarer and have been associated with the genesis of cervical lung hernia and with spontaneous pneumothorax.

In 2010, we reported for the first time the presence of the apical pleural holes (4): in a series of 750 patients operated on for upper limb hyperhidrosis by thoracoscopy using 5-mm 0° telescope, we found 5 patients (4 men and 1 woman) with an apical pleural hole, situated in all cases on the left sided. The holes were up to 5 mm in diameter, presented sharp margins, and were located laterally to the subclavian artery, and precisely on the medial side of the apical pleura. We postulate that this defect could be a possible precursor of cervical lung hernia, a rare clinical condition characterized by the protrusion of lung parenchyma in the neck beyond the limits of the musculoskeletal thorax. Usually, the protrusion of the lung into the neck is very small and asymptomatic. It is due to an anatomic defect of the underlying parietal pleura which cover the thoracic outlet and the Sibson's fascia (16). Although Yamada and co-authors described the presence of a mediastinal pleural anomaly on the left side (6), these congenital anomalies are not well known and usually go unnoticed.

Some authors find a relationship between the apical parietal pleural hole and the presence of apical lung bullae and/or blebs located in the same apical pleural area associated with the presence of pneumothorax (5,7). In fact, during thoracoscopic surgeries for spontaneous pneumothorax, Lin and co-workers incidentally found some

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apical pleural defects that they called "vascular-penetration defect" (5). Nine out of 22 patients (40.9%) with primary spontaneous pneumothorax patients were found to have these pleural abnormalities: they were small (each about 2–6 mm in diameter), round dimples with clear sharp margins, with vessels penetrating out, were found singly or in twos, but could be present up to four in number, and they were situated above the first rib. They observed that most of the blebs were also localized within the boundary of the first ribs, were complementary to the pleural holes, and found at sites on the lung abutting the apical pleural holes. Given that these apical pleura holes were crossed by vessels and the first rib circle is the place where the subclavian vessels penetrate out the thoracic cage, Lin and co-authors postulated that the colocalization of major vessels and the apical pleural holes within the first rib circle may not be coincidental. The association of these apical pleural anomalies with the pneumothorax has been explained, from an etiopathogenetic point of view, as follows: it is well known that one of the theories of the etiology of spontaneous pneumothorax is that it occurs more frequently in tall and thin subjects with a low body mass index. In these patients the negative pressure gradient is higher in the apex area of the lung and the expansion pressure is higher at the apex of the lung pulling on the pleura to form subpleural blebs (17). Patients with spontaneous pneumothorax and apical pleural defect were at the 90th percentile pf body height among the same age group. Thus, the particular aspect of the apical pleura defect (concavity), and the higher negative pressure presented in the taller patients, associated with the complementary relationship between the emphysematous-like changes and apical pleural holes, may contribute to trapping the visceral pleura causing the herniation into the pleural hole. Some abrasion and inflammatory changes may be caused by the trapped pleura which induce the emphysematous-like changes and pneumothorax (5).

In 2020, Yang and Jung (18) reported new hypothesis about the mechanism of blebs/bullae formation one of which included the presence of small holes that penetrates the parietal pleura and named "p-spot (pneumothorax-spot)". These p-spots are more visible when the lung collapse occurs in primary spontaneous pneumothorax. The authors underline the fact that it is evidence that the small bleb from the lung apex has been located in the mediastinal soft tissue layer, digging into the chest wall that has been in contact for a long time, and the presence of p-spots has been noted in the 29.7% of the cases of patients (136/458) with spontaneous pneumothorax who performed preoperative chest computed tomography. According to these authors, the p-spots, similar to the vascular-penetration defect reported by Lin *et al.* (5), are formed thanks to a mechanism of compression of the blebs in the apical pleura which push into the mediastinal soft tissue helped by an increased intrathoracic pressure accompanied by a strong intrapulmonary positive pressure (18).

More recently, Shiiya and co-authors (7), founded parietal plural small holes in the apex of the pleural while performing surgery for primary spontaneous pneumothorax. They also suggested and confirmed, as previous authors (5,17,18), that diffuse pleural porosity is associated with primary spontaneous pneumothorax. Shiiya and co-authors investigate the prevalence of parietal pleural small hole in patients with both primary and secondary pneumothorax over a wide age range evaluating the features of patients with pleural holes. Moreover, Shiiya and colleagues, compared with other authors, investigated and compared in this study the degree of chest wall flatness in patients with primary pneumothorax and pleural holes. In their experience, the incidence of these small holes was 15.2% (45/297) in patients with primary spontaneous pneumothorax and 0.5% (1/193) in patients with secondary pneumothorax. In a multivariate analysis, primary spontaneous pneumothorax was an independent predictor of pleural holes in patients with pneumothorax after adjusting for age and sex suggesting that parietal pleural small hole is nor rarely associated with primary pneumothorax. Compared to our previous work (16) and Lin *et al.*'s work (5), approximatively half of the pleural holes in Shiiya *et al.*'s study were discovered to be under the first rib, which is outside Sibson's fascia. At the light of this evidence, and considering the comparing age, Shiiya and colleagues stated that the apical holes should not be considered always as precursor of cervical lung hernia. However, we think that cervical lung hernia may have a variety of causes as etiopathogenesis including the presence of the apical pleural holes.

In contrast with Lin *et al.*'s (5) and Yang and Jung's (18) studies, Shiiya *et al.* did not find associated blebs or emphysema-like changes near the apical pleural holes invalidating the hypothesis of visceral pleural inflammation, apical pleural adhesions associated with apical pleural holes.

The flatness of the chest wall has also been evaluated for the possibility of its association with apical pleural holes. Shiiya and colleagues reported that in patients with apical pleural holes the chest wall flatness was not as severe as that

in subjects without pleural holes. This chest wall feature in patients with pleural holes still seems flatter in respect to normal controls reported in other studies, although the chest wall method of measurement was slightly different from Shiiya's group (19,20). In Shiiya *et al.*'s opinion, these features suggest that subjects with apical pleural holes may have some specific findings that leads to pneumothorax, which are different from other subjects with primary spontaneous pneumothorax.

In conclusion, different studies reported the presence of the apical pleural holes accidentally discovered during the treatment of pneumothorax or palmar hyperidrosis and the pathogenesis of which has been differently hypothesized but not fully clarified. Our previous experience led us to speculate that this apical pleural hole could be the precursor of cervical lung hernia which may develop through a defect of the parietal apical pleura and the Sibson's fascia.

Further studies with large series are necessary to investigate the underlying characteristics in subjects with these anomalous apical pleural defects definitively clarifying their etiology and role in the primary spontaneous pneumothorax.

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